When Appearances are Misleading:
Psychosocial Consequences of Facial Masking in Parkinson's Disease

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Abstract

Facial masking, the loss of expressive facial mobility in Parkinson's disease, has the potential to impact communication, emotion perception, subjective wellbeing, and social relationship functioning. This unique symptom has been studied in the context of the healthcare and casual interactions, but little is known about how it affects people who have Parkinson's in their family interactions. Since no cure for Parkinson's disease presently exists, lessening the impact of the condition is an important aspect of Parkinson's management. However, research has been missing the voices of those who experience facial masking. This gap underscores the need to deepen our understanding of this symptom and the difficulties it may create. Thus, this research project sought to understand experiences and challenges related to facial masking in Parkinson's, to transform those insights to targeted clinical tools, and to investigate the significance of masking in the psychosocial wellbeing of both people with Parkinson's and their partners or family members.

In Study 1, in-depth personal accounts were collected of nine people who had Parkinson's and their spouses or partners were separately interviewed about their experiences of facial masking. The study aimed to explore the common experiences and challenges related to facial masking, on an individual level and within close relationships. Facial masking was often falsely perceived by partners as a negative affect and wrongly interpreted to mean a lack of care or interest. The loss of expression and its misinterpretation was described as a barrier to emotional connection by some participants. Loss of the ability to physically embody one's thoughts and feelings was unsettling and distressing for both people with Parkinson's and their partners. The interviews revealed most participants had low awareness that this symptom was an aspect of Parkinson's, with nearly all indicating a wish for greater education and support around this symptom.

Study 2 involved the development and validation of two separate questionnaires that evaluated the socioemotional consequences of facial masking, one for individuals who have

Parkinson's and the other for their partner, spouse, or other family member. Questionnaire items were derived from the findings of Study 1 and a review of relevant concepts in existing literature. Psychometric properties of the questionnaires were statistically evaluated among a community sample of 80 people who had Parkinson's and 58 of their significant others. This included an item-level assessment, an exploratory investigation of the factor structure, an evaluation of the internal and test-retest reliability, and evaluation of the construct validity of the questionnaires. Results indicated the questionnaires had good reliability and initial evidence of validity. Modified (revised) versions of the questionnaires were produced based on the psychometric findings of Study 2.

Study 3 described the functioning of the participants in the psychometric study above. This study investigated the relationship between perceptions of facial masking and various psychosocial outcomes, and a study-specific measure of the impact of facial masking in Parkinson's (developed in Study 2). Ratings of facial masking were made by self-report and significant other reports, along with healthcare professional and researcher ratings based on a videotaped conversation. Healthcare professional ratings of FM were not found to be significantly related to self/significant other ratings of facial masking severity. Overall, having more negative consequences of FM was linked to poorer psychosocial outcomes, but suggested people with Parkinson's and their significant others may be affected in different ways.

Findings of this programme of research add to what is known about the consequences of facial masking in casual and healthcare provider social interactions, by examining this symptom from the perspective of those living with facial masking in Parkinson's. The results build upon and are generally consistent with the small amount of existing evidence showing that facial masking can have a detrimental impact on subjective wellbeing. The main implication of this research is that facial masking is a topic that warrants greater clinical attention and communication. The findings also call attention to the fact that facial masking is a common feature of Parkinson's, yet in New Zealand, people with the

condition and their families seem to lack knowledge about this symptom and desire greater education and support.

Dedication

To Pete, who was not able to see

the completion of this work.

It was your love and support that

made this possible.

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Contents

List of Publications Included in Thesis

Manuscripts:

Wootton, A., Starkey, N. J., & Barber, C. C. (2019). Unmoving and unmoved: experiences and consequences of impaired non-verbal expressivity in Parkinson's patients and their spouses. *Disability and Rehabilitation*, *41*(21), 2516–2527. https://doi.org/10.1080/09638288.2018.1471166

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Wootton, A., Starkey, N. J., & Barber, C. C. (2020). *Face Value: Perception of Facial Masking and the Link to Psychosocial Outcomes in Parkinson's Disease*. [Manuscript prepared for publication].

Conference and Clinical Practice Presentations:

Wootton, A., Starkey, N. J., & Barber, C. C (2014). *Facial masking: a silent killer of close relationships in Parkinson's disease*. [Poster presentation]. The New Zealand Applied Neurosciences Committee Conference, held at Auckland, New Zealand.

Wootton, A., Starkey, N. J., & Barber, C. C (2014). *Facial Masking in Parkinson's disease.* [Presentation]. The New Zealand Psychological Society Annual Conference, held at Hamilton, New Zealand.

Wootton, A. (2014). *The Mask of Parkinson's.* [Clinical Practice Presentation]. The Parkinson's New Zealand Annual Conference, held in Auckland, New Zealand.

Wootton, A. (2020). *Hypomimia: the mask of Parkinson's*. [Clinical Practice Presentation]. Parkinson's New Zealand Annual Conference, held in Wellington, New Zealand.

Educational Presentations and Publications:

Wootton, A. (2014). *Facial Masking in Parkinson's* [Patient Educational Presentation]. Parkinson's New Zealand Waikato Early Onset Support group meeting, held in Gordonton, New Zealand.

Wootton, A. (2015). *Facial Masking in Parkinson's*. [Patient Educational Presentation]. Parkinson's New Zealand Kāpiti Coast Annual Members Conference, held in Kāpiti Coast, New Zealand.

Wootton, A. & Parkinson's New Zealand (2016). Fact Sheet: What is Facial Masking? *The Parkinsonian*, *19*(1), 6–8. Wellington, New Zealand.

Wootton, A. & Parkinson's New Zealand (2020). Fact Sheet: What is Facial Masking? [online patient education resource, revised, see thesis appendices].

1. General Introduction

Facial masking (FM) is a symptom of Parkinson's disease (PD) which causes an appearance of impassiveness and unusual stillness in the face. Investigation of this unique symptom has centred on the social perceptions of unfamiliar perceivers, overlooking the perspectives of people with Parkinson's themselves and their families. Deepening our understanding of the impact of having FM could help to inform clinical practice and promote quality of life. This thesis helps to address that gap in two ways: firstly, by increasing understanding of the experiences and consequences of FM in the lives of people with Parkinson's disease; secondly, by utilising those insights to inform the development of a questionnaire to detect the socioemotional consequences of facial masking. Existing assessments of facial masking do not address related aspects of wellbeing. This thesis presents and tests an alternative approach, drawing attention to the social and emotional difficulties related to facial masking, to better inform the management of wellbeing in Parkinson's disease and lessen the impact of this condition on quality of life.

The thesis format follows the University of Waikato guidelines for the "with publication" thesis variant. This comprises traditional thesis chapters interspersed with research articles. The first two chapters introduce the topic of the thesis. Chapter 1 describes Parkinson's disease and provides background information about the disease context. Chapter 2 explains the symptom of facial masking, provides an overview of the literature mapping changes to facial expression in PD, reviews literature on the influence of facial masking for social perception, outlines the potential mechanisms involved in others' responses to people who have facial masking, and summarises the two studies which previously investigated the psychosocial consequences of masking in people with Parkinson's and their partners. A summary and identification of gaps in the current literature is also provided. Chapter 3 outlines the present doctoral project with a description of the research aims, design, and assumptions, and briefly introduces each of the studies comprising this thesis. Chapters 4–6 comprise the three separate studies. In Chapter 4,

Study 1 presents a qualitative investigation of the experience of having facial masking. Study 2 (Chapter 5) presents the development and validation of two novel questionnaires assessing FM impact. Study 3 (Chapter 6) presents an exploration of the link between facial masking and subjective wellbeing in Parkinson's disease and compares how different perceiver ratings of masking severity are associated with psychosocial outcomes. A mixed methods summary of the integrated findings is presented in Chapter 7. Lastly, general discussion of the research findings, clinical implications, and conclusions are provided in Chapter 8.

Parkinson's disease: An Increasing Burden

PD has recently been identified as the fastest growing neurological disorder, exceeding the growth of conditions like Alzheimer's disease (Dorsey et al., 2018). Across the lifespan, PD is the most common movement disorder and, together with essential tremor, is the second most common neurodegenerative disorder (Tanner & Aston, 2000). Rates of PD tend to rise sharply from middle age in studies of western populations, with estimates sitting at approximately 1% in the population aged 60 years (Nussbaum & Ellis, 2003; de Lau & Breteler, 2006) and 3–5% of the population aged 85 years or older (Fahn, 2003). PD does occur in younger age groups with approximately 4% of all PD patients showing clinically overt signs between the ages of 21–50 years (Alves et al., 2008). Incidence rates vary between studies and across countries, but a systematic review found international rates of 220 to 670 per 100,000 per year for individuals 65–84 years of age, with the risk of PD peaking at about 75 years of age (Twelves et al., 2003). In Aotearoa New Zealand (NZ), the standardised incidence and prevalence rates of PD were most recently calculated from a community database of anti-parkinsonian medication users (Pitcher et al., 2014; Pitcher et al., 2018). Based on the total NZ population in 2013, values of 31 per 100,000 per year and 220 per 100,000 per year were calculated for PD incidence and prevalence respectively (Pitcher et al., 2014; Pitcher et al., 2018). Using these rates, this would equate to just over 10,000 individuals with PD in NZ in the year 2020. However, there is a consensus that most

estimates are likely to underrepresent the true rate, and therefore burden, of Parkinson's disease. This is because many individuals may remain undiagnosed and because of diagnostic uncertainty during early stages of the condition. Early symptoms may be wrongly attributed to old age or other neurological conditions, and it is not unusual for a delay of months to years between the appearance of symptoms and clinical diagnosis (Breen et al., 2013).

Several factors contribute to the increasing social and economic burden of Parkinson's disease. In New Zealand, the growth of Parkinson's disease is partly explained by an aging population structure (Wang et al., 2012), and because PD is mostly a condition of middle to later life (Alves et al., 2008), the number of people with aging related disorders such as Parkinson's conditions is rising. Second, present therapies for Parkinson's disease have limited efficacy. Even optimal pharmacological treatment only partially alleviates symptoms, and long-term use is constrained by dosage limits and the development of medication-related complications, such as uncontrolled movements (van der Marck et al., 2009). Surgical and infusion treatment is usually only considered in a small number of cases; the benefits, and drawbacks of which are briefly discussed below. Third, the disease course is characterised by increasing disability paired with declining quality of life, yet there is limited impact on life expectancy. Factors which predict mortality in Parkinson's populations are not well understood; survival seems dependent on the individual's disease profile. For example, the survival rates for individuals diagnosed with Parkinson's before the age of 60 (Chaudhuri, et al., 2011) and for individuals with Parkinson's who have largely intact cognitive function (Bäckström et al., 2018) are often reported to be similar to the general population. Older age at diagnosis and the presence of dementia or other Parkinsonism conditions are generally associated with a reduced rate of survival relative to the general population (Macleod et al., 2014). The most common cause of death in all people with PD is typically a comorbid disorder such as pneumonia (Beyer et al., 2001; Fall et al., 2003). While Parkinson's disease itself is not fatal, it is a condition characterised by chronic and pervasive dysfunction, with most individuals becoming increasingly disabled and experiencing

significant reduction across all areas of quality of life (van der Marck et al., 2009; Williams & Litvan, 2013). The wide-ranging symptoms of PD require integrative, patient-centred approaches to routine PD care (Eggers et al., 2018). However, the provision of day-to-day support often relies on familial caregivers until the individual has high care needs. Given these unique disease factors and with the expected number of people with PD on the rise, it is important to document the broader implications of PD in the context of an individual's life.

Clinical Characteristics, Pathology, and Aetiology

Parkinson's disease (PD) is a progressive neurodegenerative disorder. Historically considered a disorder of movement, current understanding of PD also incorporates a wide range of motor and non-motor symptoms. PD falls under the umbrella of Parkinsonism, a general term for a cluster of neurological disorders characterised by movement-based dysfunction. The essential feature of this syndrome is bradykinesia, a slowness and reduced amplitude of muscular movement with a weakening of repetitive movements, along with additional features of rigidity and/or resting tremor (Bartels & Leenders, 2009; Williams & Litvan, 2013; Postuma et al., 2015). Aside from PD, Parkinsonism also encompasses corticobasal disease, multiple system atrophy, progressive supranuclear palsy, Parkinson's with Lewy bodies, and Parkinson's dementia (with the latter two conditions collectively known as Lewy body disease). Clear definition among these disorders is yet to be established, in part because there is overlap of clinical presentation and treatment options. However, when compared to other Parkinsonism disorders, PD is the most prevalent and is characterised by relatively better outcomes in treatment and prognosis. Current diagnostic criteria for PD includes the presence of at least two of the following motor symptoms: resting tremor, bradykinesia, rigidity or postural imbalance, and the absence of atypical features which may indicate another condition (Fahn, 2003; Alves et al., 2008; Jankovic, 2008). Other key features of PD include secondary motor symptoms, such as gait changes and speech difficulties, and non-motor symptoms such as autonomic dysfunction, cognitive and neurobehavioral abnormalities, sleep disorders, and sensory abnormalities.

Onset of PD is often subtle, and the course of the disease is characterised by great individual variation in symptomology and progression. Preclinical and dominant symptoms vary widely across people with the condition. There is no widely available definitive test for PD, so diagnosis typically relies upon the presence and progression of clinically overt features (Litvan et al., 2003; Bartels & Leenders, 2009). Discriminating between conditions of Parkinsonism may be not immediately possible, and it is not uncommon for initial diagnoses to be updated over the disease course. Some individuals may be diagnosed under the general term "parkinsonism" and only receive a definitive diagnosis at autopsy or through longitudinal observation of the disease course (Clough & Sethi, 2003; Alves et al., 2008). In New Zealand, a clinical diagnosis of Parkinson's involves a medical history, an interview, and a neurological examination (Neurological Foundation of New Zealand, 2013). Several additional criteria are supportive of establishing a PD diagnosis and are considered important features in discriminating among types of Parkinsonism, such as asymmetric symptom onset and a positive response to anti-Parkinsonian medications (Williams & Litvan, 2013).

The stages of PD progression may be classified using a rating system (Hoehn & Yahr, 1967). Recent revisions of the system classify five stages and two substages of the disease (Goetz et al., 2004). Progression is seen from a unilateral presentation with very minimal functional impact to bilateral presentation with increasing impairment of functionality (see Table 1). The median time between PD stages has recently been evaluated in a relatively large group of people with PD (*N* = 695, mean age in years = 65.2, male gender = 57.3%). The transition between stages 2 to 2.5 was identified as generally the longest, at a median time of 62 months, with all other stage transitions between 20–26 months. The total (median) transition time from the first to final stage was found to be 157 months or approximately 13 years (Zhao et al., 2010).

Table 1

Simplified description of clinical classification of PD staging and median time to transit between stages.

Stage:	Characteristics:	Time to transit stages
0	No signs of disease	-
1	Unilateral involvement only; minimal or no functional impairment	-
1.5	Unilateral disease	-
2	Bilateral disease, without impairment of balance	20
2.5	Mild bilateral disease	62
3	Mild to moderate bilateral disease; some postural instability; independent but needs help with activities of daily living.	25
4	Severe disability; still able to walk or stand with help of equipment	24
5	Wheelchair bound or bedridden unless aided	26

Note. PD staging descriptions simplified from Goetz CG, Poewe W, Rascol O, et al. Movement Disorder Society Task Force report on the Hoehn and Yahr staging scale: status and recommendations the Movement Disorder Society Task Force on rating scales for Parkinson's disease. *Movement disorders* 2004;19(9):1020-28.
Median transit time in months as reported by Zhao YJ, Wee HL, Chan YH, et al. Progression of Parkinson's disease as evaluated by Hoehn and Yahr stage transition times. *Movement Disorders* 2010;25(6):710-16.

The symptoms of Parkinson's disease are thought to be caused by neurodegeneration (cell death), which disrupts nigro-striatal circuits of the brain through the depletion of the neurotransmitter dopamine (Hornykiewicz, 2008). The hallmark of this condition is the selective death of cells in a region called the substantia nigra pars compacta, a component of the basal ganglia system located within the mid-brain (Less et al., 2009). The basal ganglia are involved in several brain functions but are essential to the facilitation and coordination of motor movement. In simple terms, when the decision to make a movement occurs, the inhibition of the basal ganglia on the motor system is released. This leads to excitation of the motor cortex, which then signals the muscles and allows movement (Bonnet & Houeto, 1999; Takakusaki et al., 2004). The neurotransmitter dopamine is essential to enhancing the actions of basal ganglia pathways underlying motor movement.

When dopamine is depleted in PD, the inhibition of the basal ganglia on the motor cortices is harder to release, and the net result is difficulty in initiating and coordinating movement. Put simply, declining levels of dopamine in PD reduce the ability of the brain to effectively communicate with the muscles of the body. This makes movement slower and less coordinated and sometimes causes abnormal movements like shaking (tremor) or freezing. The depletion of dopamine also has implications for non-motor functions such as cognition, emotion, behaviour, sensory perception, and autonomic processes (Chaudhuri & Quinn, 2014).

While the neurodegenerative process of Parkinson's disease is well understood, the aetiology of this degeneration has been less well-established. Researchers have generally proposed that an interaction of environmental, occupational, lifestyle factors, and a genetic susceptibility contribute to the pathogenesis of the disease. Risk factors consistently mentioned in the literature include the following: exposure to pesticides (Brown et al., 2006); head injury in early life (Taylor et al., 2016); and male gender (Picillo et al., 2017). Other variables which have limited, or conflicting evidence include exposure to environmental pollutants (heavy metals, organic solvents, magnetic fields); high levels of inflammation; comorbidity (diabetes, cancer, and other neurological conditions); high alcohol consumption; low physical fitness; and various dietary patterns or food groups (de Lau & Breteler, 2006; Wirdefeldt et al., 2011). Although a genetic contribution to the disease has been long proposed, genetic mutations are thought to account for only small proportion (<10%) of the aetiology of this condition, outside of the small minority of hereditary Parkinson's disease (Alves et al., 2008).

The Status of Therapeutic Options

No current treatment effectively prevents or halts the progression of PD. Since its introduction in the 1960s, the mainstay has been dopamine replacement therapy, which aims to increase levels of the neurotransmitter dopamine in the brain (Hornykiewicz, 2010). Despite their name, these therapies act by enhancing the synthesis of dopamine from

remaining substania nigra cells, rather than replacing it. Unfortunately, within a few years of treatment and due to the progression of the disease, the benefits of dopamine replacement therapy fade, and likelihood of unpleasant complications increases (Marsden & Parkes, 1977; Marsden, 1994). The limitations of dopamine replacement therapy led to the introduction of other treatments including catechol-O-methyltransferase (COMT) and monoamine oxidase type B (MAO-B) inhibitors, which act by extending levodopa/dopamine half-life. To minimise symptom fluctuations in response to oral pharmacotherapy, the delivery of levodopa and dopamine agonists can be improved by using an infusion pump, which continuously delivers the agents to the bloodstream. Surgical procedures also exist, such as deep brain stimulation, a form of electrical stimulation via a pacemaker-like device which can restore function of the motor system, possibly through minimising abnormal activity in mid-brain structures (Lozano et al., 2002). Although generally considered to be safe and effective, surgical procedures are typically recommended to only those patients who have responded poorly to pharmacological intervention, such as having severe side effects or fluctuations in response to medications (Lozano et al., 2002). While surgical treatment offers clinical improvement of motor symptoms in PD it does not treat the root cause, providing limited benefit for the progression of dopamine depletion (Hilker et al., 2005). Only a small number of people are approved each year for surgical treatment of Parkinson's disease in New Zealand (Neurological Foundation of New Zealand, 2013). Given present limitations on medical treatment and the chronic decline of functioning that occurs in Parkinson's, it is perhaps not surprising to find that a growing body of literature focuses on improving subjective wellbeing and quality of life in this condition.

Expressive Loss in the Parkinson's Context

To this point, Parkinson's disease has been framed as neurodegenerative movement disorder primarily caused by a dopamine deficiency. However, difficulties carrying the movements required for walking or in activities like dressing are only part of the picture. The many and varying symptoms of the disease challenge an individual's bodily agency, social

identity, and self-concept (Bramley & Eatough, 2005). Often, PD is described to imprison the individual within an increasingly unresponsive shell (Anonymous, 1999; Tickle-Degnen, Zebrowitz & Ma, 2011). Not only are people out of tune within their own bodies, but also diminished in their ability to communicate and express themselves to others due to the disease-related changes. The following sections briefly introduce the modalities of self-expression that may become impaired in PD, such as speech and facial movement.

Speech and communication deficits are common symptoms of Parkinson's, evident in up to 89% of individuals (Dashtipour et al., 2018). Verbal communication is impacted by wide ranging symptoms stemming from both the physical and cognitive production of speech (Forrest et al., 1989; Sapir et al., 1999; Dashtipour et al., 2018). Deficits in emotional expression also occur in verbal communication, namely in the impairment of emotional tone or prosody of the voice (Penner, 2001; Schröder et al., 2010). Changes to vocal prosody in PD (a loss of loudness, alternations to pitch, and a slowing of the tempo of speech) tend to be observed by others as sad or lacking emotion (Pell et al., 2006). It has been proposed that the social consequences of changes to vocal prosody are not only a difficulty in communicating intelligibly but also decreased social-linguistic competence and a tendency for listeners to perceive the communication of people with PD in misleading or detrimental ways (Pell et al., 2006). Further to this, one the most impactful aspects about having a speech disorder in PD was the effect on social participation and family connections, (Miller et al., 2006). Miller and colleagues (2006) also found the social impacts of changes to speech occurred before speech intelligibility was overtly compromised. This suggests it is clinically important to address disease related changes with respect to their impact for psychosocial wellbeing, and this might require earlier intervention than waiting until physical functioning is impaired (Miller et al., 2006).

Other abnormalities of bodily movement occur (including but not limited to tremor, freezing, dyskinesia, and dystonia) and may contribute to a visible physical difference, marking out individuals who have PD in ways that tend to be negatively interpreted by others. Abnormal moments in the muscles of the body in PD also occur in the face. The

rigidity, slowness, and decreased coordination seen in the movement of the body similarly affects facial musculature, causing a reduction of movement and therefore emotional expression in the face.

Facial Expressive loss in other clinical conditions

Research has highlighted that the expression of emotion is vital to effective communication, crucial to the ability to partake in social interactions, and a fundamental aspect in determining subjective wellbeing (Ishii et al., 2011; Ishii et al., 2018), and has explored the role of facial expressive loss in other clinical conditions. Deficits in facial movement and expression occur in number of medical and psychiatric conditions. This includes congenital or acquired facial paralysis where one side or all the facial muscles lose the ability to move (Valls-Solé & Montero, 2003); and traumatic brain injury, where there can be a reduction in the expression of emotions (Kupferberg et al., 2001; de Sousa et al., 2011; McDonald et al., 2011). Facial expressive deficits may also occur in psychological disorders such as depression where the experience and expression of emotion may become flattened (Jaeger et al., 1986; Gaebel & Wölwer, 1992; Sloan et al., 1997); and in schizophrenia, where the experience of emotion may become dissociated from displays of expression or emotion on the face (Bleuler, 1911; Andreasen, 1982; Gelber et al., 2004; Gur et al., 2006; Kohler et al., 2008). Facial expressive loss in PD is set apart from these conditions because FM is gradually acquired, progressive, and because there is often simultaneous impairment of the other channels of communication such as speech and bodily gesture (as outlined in the section above). This uniqueness also means knowledge drawn from other clinical populations about the consequences and treatment of facial expressive loss may not be universally applicable to FM in PD.

Nevertheless, literature in other clinical populations recognises the significance of facial expressive loss and encourages awareness of the broader implications in social and emotional functioning. In brain injury, the reduction of expressivity is linked to deficits in empathy (de Sousa et al., 2011) and emotional mimicry processes (McDonald et al., 2011).

In congenital facial paralysis, decreased self-perception of the ability to communicate emotion has been associated with reductions in quality of life (Coulson et al., 2004). There is evidence of the negative influence of facial paralysis on subjective emotional well-being and the ability to maintain relationships, emotion recognition abilities of perceivers; and with respect to issues and interventions for communication (May & Schaitkin, 2000; Bogart et al., 2012; Bogart et al., 2014; Bogart & Tickle-Degnen, 2015; Michael et al., 2015). In acquired facial palsy, a positive association between the altered or diminished capacity for expression and greater psychological distress has been shown (Fu et al., 2011). In facial neuromotor disorders, a specific impairment in the ability to smile has been associated with greater levels of depressive symptoms (Van Swearingen et al., 1999).

The social and psychological dimensions of facial expressive dysfunction in PD have not received similar attention in scientific literature. This is perhaps because the traditional focus of PD treatment has been on the physical symptoms which lead to difficulties in performing activities of daily living. Indeed, the characteristics healthcare providers focus clinical attention on in PD often differ from patient reports of the most bothersome symptoms. In one study, dressing, ambulating, and cognitive difficulties associated with motor sequencing were considered by experts to be characteristic problems of Parkinson's, but patients themselves reported that such issues concerned them less than cognitive and psychosocial symptoms of the condition (Abudi et al., 1997). The focus of clinical communication and treatment in PD may have changed in the last 20 years, yet symptoms like FM, which may be less physically debilitating but take a toll on subjective wellbeing, have received far less attention in existing research.

2. Facial Masking in Parkinson's disease

Clinical Features & Occurrence

Facial masking is variously called hypomimia, amimia, facial bradykinesia, masked facies, or more colloquially as "the Parkinson's mask". This thesis uses the term facial masking or FM. From a medical perspective, FM is defined as the loss or reduction of facial expression. From patients' perspective, FM is a reduced ability to express one's emotions, thoughts, and characteristic behaviours on their faces (Lyons et al., 2004). The net result being the individual seems increasingly unresponsive, and therefore uninterested, in the environment around them (Bologna et al., 2013). Clinical staging instruments define FM as a loss of facial emotional expressivity which is manifested by a decreased blink frequency, less spontaneous smiling, and the parting of lips when the mouth is at rest (Goetz et al., 2008). Neurophysiological studies of the face in Parkinson's disease have also identified various facial folds which become flattened around the nose, mouth, and eyes (Bandini et al., 2017).

Disease-related changes to positive expressions, as well as an overall reduction of expressivity, seem to play a significant role in FM in PD. Video analysis of behavioural cues during naturalistic interaction in people with PD and matched cardiac disease controls revealed individuals with PD showed a majority of smiles without a cheek raise, known as phoney smiles, with very few genuine smiles (Pitcairn et al., 1990).The Parkinson's group also showed very few other emotional expressions, while the cardiac group showed a range of different expressions (Pitcairn et al., 1990). Posed expressions of happiness have also been studied and are shown to be less recognisable and lack a cheek raise (Simons et al., 2004), and spontaneous expressive reactions to humorous stimuli are also reduced in frequency in people who have PD compared to healthy controls (Katsikitis & Pilowsky, 1991).

Despite being a distinctive feature of PD, just a few studies have formally measured the occurrence of FM. In one Swedish longitudinal study, various orofacial symptoms in PD

were investigated from routine care visits (Fereshtehnejad et al., 2017). FM was measured by the Unified Parkinson's Disease Rating Scale (MDS-UPDRS) (Goetz et al., 2008), with FM recorded in approximately 37% of the participants at baseline, rising sharply to around 92% of participants at follow-up, where the average follow-up duration was 4.2 years (Fereshtehnejad et al., 2017). Another recent study provided a description of 89 people who had PD who were also clinically evaluated using the MDS-UPDRS, finding some degree of FM present in 70% of the sample cohort at the point of testing (Ricciardi et al., 2020). For comparison, other characteristic symptoms of PD occur at similar rates. Resting tremor has been observed in 75% people with PD throughout the stages of the disease (Hughes et al., 1993), and speech difficulties are found in 89% (Dashtipour et al., 2018). This suggests FM, while not as well-known or noticeable as other symptoms of PD, is just as ubiquitous.

Previous FM research in PD has focused on three main areas of investigation. First, early papers typically turned attention to defining facial movement deficits and their underlying neural mechanisms. Second, the implications of FM were examined for general social functioning, namely in the context of casual and healthcare interactions. Third, a small number of recent studies have examined the consequences of FM from patient-centred perspectives. The sections below summarise key findings from each area of existing research and highlight some of the gaps in present knowledge of FM.

Mapping Facial Masking

Early investigations of FM concerned the differential impairment of spontaneous versus voluntary facial movements and the related discussion of the neural bases underpinning facial motor control. While there was a consensus that spontaneous expressions were impaired in Parkinson's, some disagreement existed about the impact on voluntary expressions. This debate originates from theories about the neural bases of the motor system. A classic review of the neuropsychology of facial expression suggested the impairment of basal ganglia function underlies the reduced spontaneous expressivity observed in Parkinson's disease (Rinn, 1984). In contrast, posed (voluntary) expressions

were thought to originate from cortical regions, with voluntary facial movement believed to remain intact. Initially, evidence seem to give weight to the dissociation of movement systems. In one such study, the intensity and frequency of posed and spontaneous expressions of PD patients and healthy age-matched controls were compared while watching emotionally evocative stimuli. The people with Parkinson's had fewer overall spontaneous facial reactions to the stimuli than controls (Smith et al., 1996). In contrast, the posed expressions of the participants seemed to remain intact (Smith et al., 1996). Later research put forward that volitional and spontaneous expressions might be differentially impaired. More specifically, the individuals with Parkinson's were found to have greater difficulty concealing or intensifying their negative expressions and a slightly increased difficulty in intentionally mimicking facial movements than the control group (Simons et al., 2003).

The type of relationship between the person with masking and the perceiver may also impact expressivity. When people with PD were recorded speaking with a researcher and then with their spouse, participants displayed greater expressivity when interacting with the researcher (an attentive but unfamiliar person) than when secretly observed in conversation with their spouse (Simons et al., 2004). There was some inconsistency in the findings, however, as the posed expressions of the same people with Parkinson's (when explicitly instructed) were found to be markedly impaired. Although it is possible that voluntary expressive ability varies by social context, the researchers suggested this finding could be better explained by the heightening of emotion when an amicable but unknown individual is present, which prompted greater expressivity from the participants with PD (Simons et al., 2004). This suggests greater expressive loss occurs in primary social relationships, rather than with unfamiliar social partners. Thus, it could be proposed that any consequences of FM are amplified in private social contexts, such as with family and close friends.

At present, the scientific literature seems to have reached consensus that marked impairment occurs to spontaneous facial expression, and while impairment to voluntary

facial expression does take place, it is typically less severe than that of spontaneously arising facial movements. Several studies have contributed greater detail to the understanding of voluntary facial expressive impairment in Parkinson's disease. Compared to healthy controls, the voluntarily induced (posed) facial expressions of Parkinson's patients were reduced in speed and size (Bowers et al., 2006). Computer-based analyses of posed smiles found these expressions have a lower speed and intensity in individuals with Parkinson's compared to healthy controls, and that posed expressions were not improved by dopamine replacement therapy (Marsili et al., 2014). Currently, it is proposed the loss of fluidity and spontaneity of all movement (controlled by the basal ganglia system) has a negative influence on voluntary movement, as well as an evident impairment of spontaneous movement (Rinn, 2007). Two major theories have been proposed to explain a basal ganglia-controlled deficit in expressive movement. One theory proposes an impairment in the activation of motor program circuitry for movement such as facial expressions. Both selected and non-selected motor programs become suppressed, along with an impairment of the timing of activation of different programs (Nambu, 2005; Bowers et al., 2006).The second theory describes a corruption of motor programs responsible for movement of facial muscles and the resultant facial expressions (Bowers et al., 2006)

More recent trends in FM research explore techniques for recording the extent of facial expressive loss and identifying the presence of FM. Computer and video-based methods have utilised both objective assessments and subjectively coded rating systems to analyse facial behaviours (Wu et al., 2014; Gunnery et al., 2017). This is discussed further in Study 3, so will not be elaborated upon here.

Impact of Facial Masking on Perceiver Impressions

The impressions formed by perceivers when interacting or viewing a person with FM show a consistent negative bias. Investigations of how people with FM are socially perceived suggest that inaccurate judgements are made of character, mood, or mental state (Lyons et al., 2004; Tickle-Degnen et al., 2011; Friedman, 2017). In one early observation of FM,

clinicians were warned the person with PD may appear to have lost feeling or intelligence because they do not seem engaged in conversation, and that true character may be concealed by an outward impression of coldness and indifference (Monrad-Krohn, 1957). Recent evidence shows perceivers evaluate individuals with FM in an unfavourable light, describing them as cold, moody, unfeeling, avoidant, or even intellectually impaired (Monrad-Krohn, 1957; Pentland et al., 1987; Abudi et al., 1997; Chiong-Rivero et al., 2011; Hemmesch, 2014). For the most part, research exploring this impression bias has concentrated on the viewpoints of unfamiliar perceivers. For example, when strangers judged the social desirability of people with Parkinson's and healthy age-matched controls from videotaped interview excerpts, people with Parkinson's were rated as less involved, interested, intelligent, optimistic, attentive, and physically attractive (Schwartz & Pell, 2017).

Unfamiliar perceivers are not the only group susceptible to misinterpreting FM as feelings or attitude. Even healthcare professionals make more negative judgements of people with FM. When clinicians were shown silent videotapes of people with heart disease and people with Parkinson's disease, judgements of the individuals with Parkinson's were negatively biased (Pentland et al., 1987). Clinicians had unfavourable evaluations of the people with Parkinson's' intellect, affect and personality traits, despite there being no measured difference between the Parkinson's and Cardiac groups. When 19 speech therapy students were shown videos of four people with Parkinson's and four people with ischaemic heart disease, impressions were measured using visual analogue scales (Pentland et al., 1988). Both groups demonstrated irregularities on standard psychological testing, but compared to the cardiac group, the PD group were rated as "more anxious, hostile, suspicious, depressed, bored and tense than the controls; they seemed less intelligent, more introverted and passive and looked as if they enjoyed and maintained their part of the conversation less well" (Pentland et al., 1988, pg. 31). Even for experienced healthcare practitioners with greater knowledge of PD, it may be difficult to override the usage of facial expressions to form impressions of personality. A more recent study compared personality ratings of expert and student rehabilitation practitioners who viewed videos of people with

Parkinson's who had varying levels of expressive dysfunction (Lyons et al., 2004). Unsurprisingly, the students were reported to form particularly inaccurate judgements of personality as FM increased, but both students and experts rated individuals with higher masking less positively, especially in interpersonal and affective domains (Lyons et al., 2004). The authors concluded that even experienced practitioners may be prone to using behavioural cues associated with Parkinson's symptomology to make judgements of personality or other individual characteristics (Lyons et al., 2004).

The greater the degree of expressive dysfunction, the more negative impressions of an individual with FM tend to become. For example, healthcare professionals evaluated individuals with higher levels of FM as less socio-emotionally competent and less likeable than those with mild FM (Lyons et al., 2004; Tickle-Degnen et al., 2011). This finding has also been replicated with naive observers who were not familiar with the person they were watching. When strangers viewed video clips (with filtered verbal content) of people with FM, they were less interested in relationships with individuals who had a higher severity of FM (Hemmesch, 2014). Greater masking has also been associated with impressions of less social supportiveness (Hemmesch et al., 2009).

Theoretical Explanations of Facial Masking

The following sections summarise the social and cognitive mechanisms currently thought to underlie the effects of FM. Two broad areas of research that may help to explain how FM is interpreted with reference to emotional perception and social interactions.

Interaction with Social and Cultural Norms

The impact of FM in PD may be greater when it contradicts social or cultural expectations of behaviour. Researchers have suggested that FM could have greater influence on perceivers' impressions of women who have Parkinson's compared to men who have Parkinson's. In Western societies, women are expected to be more socially expressive than men (Fischer & Manstead, 2008; Hemmesch et al., 2009), so when expressivity is disrupted by Parkinson's it may put women with PD at greater risk of being misjudged by

perceivers for the lack of expression (Hemmesch et al., 2009). Gender bias in healthcare practitioner's impressions of people with PD has also been evidenced, with more negative impressions formed of women with FM than men with FM (Tickle-Degnen et al., 2011). There is limited evidence that the impression bias resulting from FM extends to different cultures. When experienced Taiwanese and American healthcare professionals were shown speech filtered video clips of people with PD, practitioners from both countries judged individuals with higher levels of masking in ways that were less favourable those with lower masking (Tickle-Degnen et al., 2011). As pointed out by Tickle-Degnen and Lyons (2004), more research is needed to understand how practitioners, or any social partner, can better attune to markers of personality and mental state in individuals with Parkinson's.

Neuropsychological Mechanisms

Few studies have explored the neuropsychological basis of disruptions to social functioning that result from facial masking in PD. Various theories of emotion may help to explain the possible influence of FM for human interaction. The following section summarise these theories in relation to FM for perceivers and then for people with PD, as originally outlined in the work of Argaud and colleagues (2018).

The apparent pervasiveness of the FM impression bias, even in groups such as doctors who are aware facial behaviours are invalid cues of mood or personality in PD, may occur because the brain processes emotional signals automatically and without intention or conscious awareness of doing so (Lakin & Chartrand, 2003; Lakin, 2006; Lakin et al., 2008). In laboratory conditions, inferences made about social characteristics can be performed as quickly as 100 milliseconds (Willis & Todorov, 2006).

One explanation for why FM leads others to come to the wrong conclusions about the thoughts, feelings, and attitudes of someone with FM might be the impact of the loss of mimicry for the perceiver. Humans rapidly and automatically synchronise facial activity with that of people around us, often without conscious awareness; this activation can be so subtle that it is not easily visible (Dimberg, 1982). The process of invoking in ourselves what we

see in others aids empathy and enables understanding of other mental states, actions, and intentions (Carr et al., 2003). When a mechanical or chemical block of facial musculature (and therefore facial expressions) is performed experimentally in healthy individuals, emotion recognition in those individuals becomes temporarily compromised (Oberman et al., 2007; Neal & Chartrand, 2011; Ponari et al., 2012). It has previously been shown that when people with Parkinson's are less able to display expressions, it can make them appear cold or uncaring to perceivers (Smith et al., 1996; Brozgold et al., 1998). This may be because signals that would be mimicked by and facilitate emotion understanding for the perceiver are blocked by the facial masking of the person with PD. It might also be that the face of someone with FM does not synchronise (mimic) emotion of the perceiver during social interactions, cutting off feedback of the perceiver's own emotion in the face of the person with FM.

Perceivers may judge the emotional or mental state of people with FM as negative because socially reinforcing expressions, like making a smile, seem to be selectively affected in PD. People with PD are demonstrated to be able to reproduce a frowning expression to some degree but are shown to have greater difficulty mimicking the smile of another person (Livingstone et al., 2016). The ability of people who have PD to mimic some expressions (e.g., frowning) but not others (e.g., smiling) may reflect that areas of the face are thought be somewhat neurologically independent but tend to be perceived as an overall signal by others (Gunnery et al., 2017).

When we see the emotional displays of other people, we respond with subjective internal experience as well as external expressive action (Dimberg & Thunberg, 2012). Emotion contagion is a process where emotional states of others are mirrored in our own felt experience (Hatfield et al., 1993). Although mimicry and contagion processes are inter-twined, mimicry is defined by a matching of external facial displays, and contagion is defined as the internal mirroring of felt experience. Emotion contagion is thought to be an automatic process which facilitates the felt experience of emotion (Hess & Blairy, 2001). It is thought that facial expressive actions may also amplify the subjective experience of emotion, known

as the facial feedback hypothesis (Buck, 1980; Adelmann & Zajonc, 1989). When perceiver's own internal mirroring of felt emotion or expressive actions are blocked by FM, they may have greater difficulty accurately recognising the emotion of the person with PD.

With respect to the experience of people with PD, it is much less clear how emotion simulation processes such as emotion mimicry, emotion contagion, and facial affective feedback are influenced in people who experience FM and what this could mean. There is evidence that people with Parkinson's also have difficulty recognising the emotions of others, with some speculating deficits in emotion perception could be associated with reduced mimicry (Peron et al., 2012; Ricciardi et al., 2015). Whether a mimicry deficit in PD is selective for certain emotions, or more generalised, has been debated. One meta-analysis suggested the recognition of negative emotions is more greatly impaired, compared to positive emotions (Gray & Tickle-Degnen, 2010). However, another recent study found a decrease in facial mimicry for mainly positive expressions that was associated with poorer recognition of positive and neutral expressions (Argaud et al., 2016). A second review pointed out that the inconsistencies in these findings may be related to variations in methodology, stimuli, or analytic procedures, such as using a restricted range of emotions or the way emotions were classified (Argaud et al., 2018). On the other hand, there is evidence in non-clinical populations that mimicry of only minimal cues is necessary to predict the internal state or behaviour of others (Baron-Cohen et al., 1997), and emotion recognition is also found to be intact in conditions where expressive mimicry is absent from birth, such as Moebius syndrome (Rives Bogart & Matsumoto, 2010).

Another factor that may influence Parkinson's disease patients and their perceptions of emotions in others is the presence of affective disorders. Depression (Cummings, 1992), apathy (Pluck & Brown, 2002), alexithymia (Assogna et al., 2012), anxiety (Latoo et al., 2013), and involuntary emotional expressive disorder (Latoo et al., 2013) can occur in Parkinson's. Along with the blocking of emotion simulation processes by FM, all of these disorders have a potentially separate or overlapping influence on how people with PD see others, and how they are perceived by others. In one study, for example, emotion

recognition deficits in PD were linked to greater levels of interpersonal distress (Clark et al., 2008). Similar findings have been observed in non-PD populations where having depression delays the processing of sad emotion information and results in a bias towards interpreting neutral faces as negative (Gollan et al., 2008). However, as noted by Argaud et al., (2018) the nature and direction of the link between depression with emotion recognition in people who have PD is currently unclear.

In summary, FM may lead other people to draw the wrong conclusions about an individual with FM's thoughts, feelings, or attitude because it hinders the usual signals which facilitate socioemotional perception in the perceiver. Two ways FM may do this are by blocking perceivers' internal mirroring of emotional experience or action, and by cutting off perceivers from receiving feedback of their own emotion in the face of someone who has PD. In people who have FM, socioemotional functioning may become compromised through the PD-related dysfunction of cognitive systems underlying emotion perception, together with or in addition to FM dampening important mechanisms of emotion simulation (Argaud et al., 2018). Where FM potentially lessens the ability to recognise and respond to the feelings of others, for people with PD and others who perceive them, it seems reasonable to assume the quality of social interactions and relationships could become compromised.

Living with Facial Masking

Relatively few studies have explored the consequences of having FM, investigated the impact of FM on social perception in primary social relationships, or explored the link between FM and wellbeing for people who have PD and those who share closely in their lives. During the planning of this thesis no research focused on living with FM for people with PD and their family relationships had been published.

Research on broader themes of the PD experience offered the first glimpses of the social and psychological consequences of FM. In a study exploring personal accounts of the impact of PD, focus groups and one-on-one interviews were conducted with PD patients (*n* = 47) and their caregivers (*n* = 15) recruited from four neurology clinics across three American

cities. Transcripts were analysed to deduce themes, using a cutting-and-sorting approach to organise and code conceptually related segments of text. One participant was noted to speak about the deep effect of FM on their personal relationships:

> "It was really hard on her in the beginning because I had symptoms, you know, a few years before and I had the mask face and everything, and ... I'd throw water in my face because I would just be so stiff in that, and evidently, she thought that I was losing interest in her, and once I got the Sinemet and we found out what happened and all the dystonia went away, you know, she cried because she thought I didn't love her anymore. She'd say something and I'd ... be smiling at her, but she couldn't really see it". (Chiong-Rivero et al., 2011, p. 61)

Perceivers may mistake the lack of expression for emotional detachment of the person they knew and loved, leading to a state of increasing alienation for both (Chiong-Rivero et al., 2011). The researchers further proposed that the loss of facial movement was emblematic of the greater PD experience as it came to signify the sense of imprisonment within an unresponsive body that characterises the disease and the reality of having an utterly life-changing condition (Chiong-Rivero et al., 2011).

Two recently published studies have investigated the connection between FM and wellbeing. The relationship between FM with social life and relationship quality was examined in one study of 40 American PD patients and their care partners, recruited from a specialist clinic and a community sample across the Boston region (Gunnery et al., 2016). It was not clear how care partners were defined, for example, whether they were also in a romantic relationship. PD and care partner participants separately rated facial masking severity by estimating the general expressivity the face using the following question: ""In general, how much difficulty does your partner have showing expression (emotion) in their face" (Gunnery et al., 2016, pg. 3). Ratings of expressive difficulty were made on a five-point scale where 1= no difficulty and 5= very severe difficulty. They also completed measures to assess social rejection (as a composite of perceived social isolation and perceived stigma in how others treat them), enjoyment of the relationship, and depression. Data were analysed

using Pearson correlations to investigate the relationships among facial masking with social wellbeing variables, and multiple linear regression was used to test whether reports of expressive difficulty predicted social rejection and partner enjoyment, independent of depression, in people with PD and their care partners.

No mean difference was found between PD participants and care partner participants ratings of expressive difficulty (t (39) = 1.00, p = .32), however, no statistically significant correlation was found between PD and care partner ratings (Gunnery et al., 2016). In care partners, higher ratings of expressive difficulty were found to be significantly correlated with less enjoyment of the relationship (r = −.55, p < .001) and greater perceptions of social rejection (r = .35, p < .05), care partner enjoyment was predicted by care partner's perceptions of the degree of expressive difficulty, but no associations were found with PD enjoyment (Gunnery et al., 2016). The relationship between greater expressive difficulty and reduced enjoyment of relationship was also found to be independent of depression. With respect to the PD participants, a significant positive correlation was found between expressive difficulty and social rejection (r = .41, p < .05), but no association was found between expressive difficulty and relationship enjoyment in the participants with PD (Gunnery et al., 2016). Depression in the PD participants was also found to have significant large correlations with their own ratings of expressive difficulty (r = .56, p < .001) and with social rejection (r = .70, p < .001) (Gunnery et al., 2016). Findings of the regression analysis showed that depression, but not expressive difficulty, predicted social rejection in PD participants. The latter is an interesting finding because it could be expected that the more expressive difficulty someone has, the more difficult socioemotional interactions could become and the less they are able to understand and share in emotions of the other, leading to decreased enjoyment in their interactions. However, that is not what Gunnery and colleagues found (2016). They proposed one explanation for this finding may be disease-related deficits known to occur in the emotion recognition abilities of people with Parkinson's (Gray & Tickle-Degnen, 2010), which means they do not accurately perceive the care

partner's response to the lack of expression, in turn sheltering their perceptions of the relationship.

Although self-reported ratings of FM by participants with PD and care partners were shown to be similar in the Gunnery and colleagues' study (2016), the ratings did not have a statistically significant correlation and the accuracy of ratings of expressive difficulty were not assessed. While there is limited evidence that people with PD are aware of their expressivity deficits, further investigation of this topic might to help explain the differences seen in the outcomes of expressive impairment for people with PD compared to their partners. It seems pertinent to evaluate the accuracy of self/partner ratings of FM severity against expert clinical evaluation or other measures of expressive ability, and to assess the level of agreement (whether ratings were concordant and where there are differences) within PD and care partner dyads. The relative lack of negative outcomes for people with PD reported by Gunnery and colleagues (2016) is puzzling, when considering qualitative evidence that proposes FM can inhibit healthy engagement in social life and relationships for PwP (Chiong-Rivero et al., 2011) and may have negative outcomes for psychological health, such as emotional distress or embarrassment about facial appearance (Abudi et al., 1997). Even if relationship enjoyment and social rejection are not affected by FM for people who have PD, it remains unknown what impact FM might have on other aspects of mental wellbeing such as anxiety, stress, and coping, or the impact on other aspects of social relationship functioning such as communication, affection, closeness, or social supportiveness. Related to this, it would be interesting to further explore the separate and shared ways that FM potentially influences the wellbeing of people who have PD and those closest to them.

In another study of FM and wellbeing, stigma and gender were explored as potential mediators of the relationship between FM and decreased quality of life (QOL) in people who have Parkinson's disease (Ma et al., 2019). Depression was also examined as a separate (parallel) mediator of the relationship between FM and QOL, because it is thought to be linked with the ability to show expression in the face (Girard et al., 2014) and linked with worse QOL outcomes in chronic illness (Earnshaw & Quinn, 2012). Experiences of stigma

have been long associated with having PD (Nijhof, 1995; Bramley & Eatough, 2005; Simpson et al., 2013). A stigmatised person is one who feels different from what is normal or expected, and as such, may feel marked out, tainted, discounted, or reduced (Maffoni et al., 2017). Ma and colleagues (2019) examined the relationship between FM, stigma, gender and quality of life in 90 Americans with Parkinson's, drawing from data of an ongoing longitudinal investigation of social self-management in PD (Tickle-Degnen et al., 2014). Participants were asked to complete a series of self-report questionnaires. Facial masking severity was assessed by asking participants to make a global self-rating of the level of facial expressive difficulty using the single item earlier reported in Gunnery et al., (2016), where a rating of 1 = no difficulty and 5 = very severe difficulty. Self-ratings were compared with clinical evaluation of FM severity on the MDS-UPDRS and found to have a small statistically significant correlation (r = .22, $p < .05$). Stigma was measured using the Stigma Scale for Chronic Illness (SSCI), a self-report questionnaire that includes assessment of felt and enacted stigma. Symptoms of depression were measured using the Geriatric Depression Scale (GDS) and quality of life was measured using the Parkinson's disease Questionnaire (PDQ-39); both are self-report measures. People with PD who reported greater expressive difficulty also tended to report more feelings of stigma. This association predicted lower levels of quality of life and was stronger when the gender of the person with PD was female. Depression was also found to mediate the impact of masking on quality of life, but this relationship was weaker than that of stigma, and the stigma pathway remained when controlling for depression. For people who have PD, this indicates that greater perceptions of stigma arise when the impassive appearance of FM breaks implicit expectations of behaviour during social interaction. Ma and colleagues (2019) proposed that when appropriate facial behaviours cannot be performed, it elicits unfavourable reactions from others by diverging from expected norms of facial behaviour or is seen as a defect. People with FM who perceive greater levels of stigma may internalise a sense of defectiveness and experience emotional distress (Ma et al., 2019). However, the role that greater perceptions of stigma play in the primary social relationships of people with PD, such as those with close

family members and friends, remains unclear.

Ma and colleagues (2019) and Gunnery and colleagues (2016) findings give a preliminary indication of some of the impacts masking may have on emotional and social wellbeing for people who have PD and for their family members. As these two studies were published after this thesis began, the present research methods and materials were not selected to build upon their work. However, the findings of the present thesis do add to what is now known about FM by further exploring the consequences of FM in the context of the close social relationships in the home.

Clinical Evaluation and Management of Facial Masking

At the time of planning this thesis around 2014, scientific literature and educational resources regarding the clinical management of FM were lacking. Patient health education was scarce and characterised by medical interpretations of FM, with information often limited to short physical descriptions and little discussion of the impact of this symptom for daily life. More recently, a growing appreciation of this symptom and broader implications is seen. A simple Google search presently reveals that several PD support organisations include educational resources about FM, how it may be misread by others, and the possible impacts for families.

Despite the progress in online information, very few clinical evaluation tools include FM. Clinical staging instruments include measurement of FM severity (such as the Movement Disorder Society's Unified Parkinson's Disease Rating Scale; Goetz et al., 2008) but do not include any questions to assess FM-related psychosocial dysfunction. PD-specific quality of life (QOL) instruments exist, but also do not include questions to assess FM impact. For example, the PDQ-39, a commonly used measure of QOL in PD, includes three questions about social functioning, but none refer to the loss of facial expression (Marinus et al., 2002). It is noted, however, that QOL instruments may still indirectly capture FM impact in PD, as they include domains potentially relevant to FM-related difficulties, such as relationship quality or communication problems.

A third area where FM is little addressed in the literature is rehabilitation. Parkinson's rehabilitation programs are typically comprised of speech, physical and occupational therapy, delivered in a group or individual format. Standardised treatment systems include the Lee Silverman Voice Treatment Programs, which are intended to help improve communication and mobility in activities of daily living (Ramig et al., 1988; Ramig et al., 1995; Farley & Koshland, 2005). Although physiotherapeutic programs are usually targeted at general mobility rather than FM per se, evidence demonstrates they may help protect and promote facial movement, at least in the short term (Dumer et al., 2014; Ricciardi et al., 2020) .Whether treatment with medication alleviates FM is also unclear as some studies show medication has little benefit on the ability to pose expressions (Marsili et al., 2014), yet others suggest overall masking severity is improved with medication (Ricciardi et al., 2020). Psychological approaches to managing FM are not reported in the published literature.

Summary & Gaps in Existing Literature

In summary, a small but growing body of literature has examined FM. Research has focused on mapping the facial motor deficits manifested in PD and has recognised the impact of FM for casual and healthcare interactions. Yet there is still much to be understood about FM. Little is known about the consequences of FM for people with Parkinson's themselves. For example, how do people who have FM and their significant others perceive, interpret, and respond to the loss of facial expression? In the scientific literature, it remains unclear whether the negativity bias shown to impact the impressions of strangers and healthcare professionals also generalises to primary social relationships, such as those with close family and friends. If unfamiliar perceivers tend to form automatic and inaccurate impressions of people with masking, would spouses or family members also have difficulty correctly detecting what their loved one is thinking and feeling? Alternatively, could the heightened familiarity between individuals in close relationships override the impression bias and any detrimental effects of facial masking? Additionally, despite initial evidence of a link between FM and psychosocial wellbeing in PD, the consequences of having facial masking

have been scarcely explored. For example, little is known about the possible factors associated with better or worse adjustment to this symptom, or whether facial masking affects people with PD and their significant others in distinct or overlapping ways. If FM has the potential to shrink vital social connections in the context of a chronic and debilitating illness, it seems pertinent to explore the social and psychological consequences of FM in PD. It is these unanswered questions which guide the focus of the present thesis.

3. Research Outline

General Aims

The overall aims of this programme of research were twofold: to better understand lived experiences of FM from perspectives of people who have PD and their significant others, and to assess the link between FM and psychosocial wellbeing in PD. Enhancing understanding of FM was achieved through gathering personal accounts of having FM in daily life and by examining the association between FM and subjective wellbeing in PD.

Until a cure for Parkinson's disease or an effective treatment for FM is found, it is imperative to find ways to help people and their families cope with the condition. Thus, the approach adopted in this research forms part of a growing trend that explores social participation and emotional wellbeing in PD (Winter et al., 2010). A focus on social and psychological wellbeing is particularly vital in PD because these domains have been found to play a larger role in patient's perceptions of health and wellbeing than physical symptoms (Abudi et al., 1997; Chrischilles et al., 2002).

The novel contribution of the present programme of research lies assessing the impact of FM for PD patients and their families, in building knowledge of the link between FM and wellbeing, and in developing targeted clinical tools that provides opportunities for dialogue about FM and the management of this symptom in day-to-day life. The broad intent of this thesis was shaped by pragmatic and applied approaches, with the research questions and methodology driven by a wish to have the findings be of value to people who experience FM and to inform professional practice.

Research Design

This research comprises a series of three studies investigating FM in people who have Parkinson's disease and individuals that are part of their primary social relationships (partner/spouse, adult family members or close friends). A summary of the research design and interconnection with the thesis format is provided in Figure 1, adapted from Berman

(2017). Broadly speaking, the research design can be divided into four phases (Creswell, Plano Clark et al., 2003):

- Qualitative data collection and analysis
- Development of a quantitative instrument and identification of variables for analysis
- Quantitative data collection and analysis
- Joint interpretation of the findings

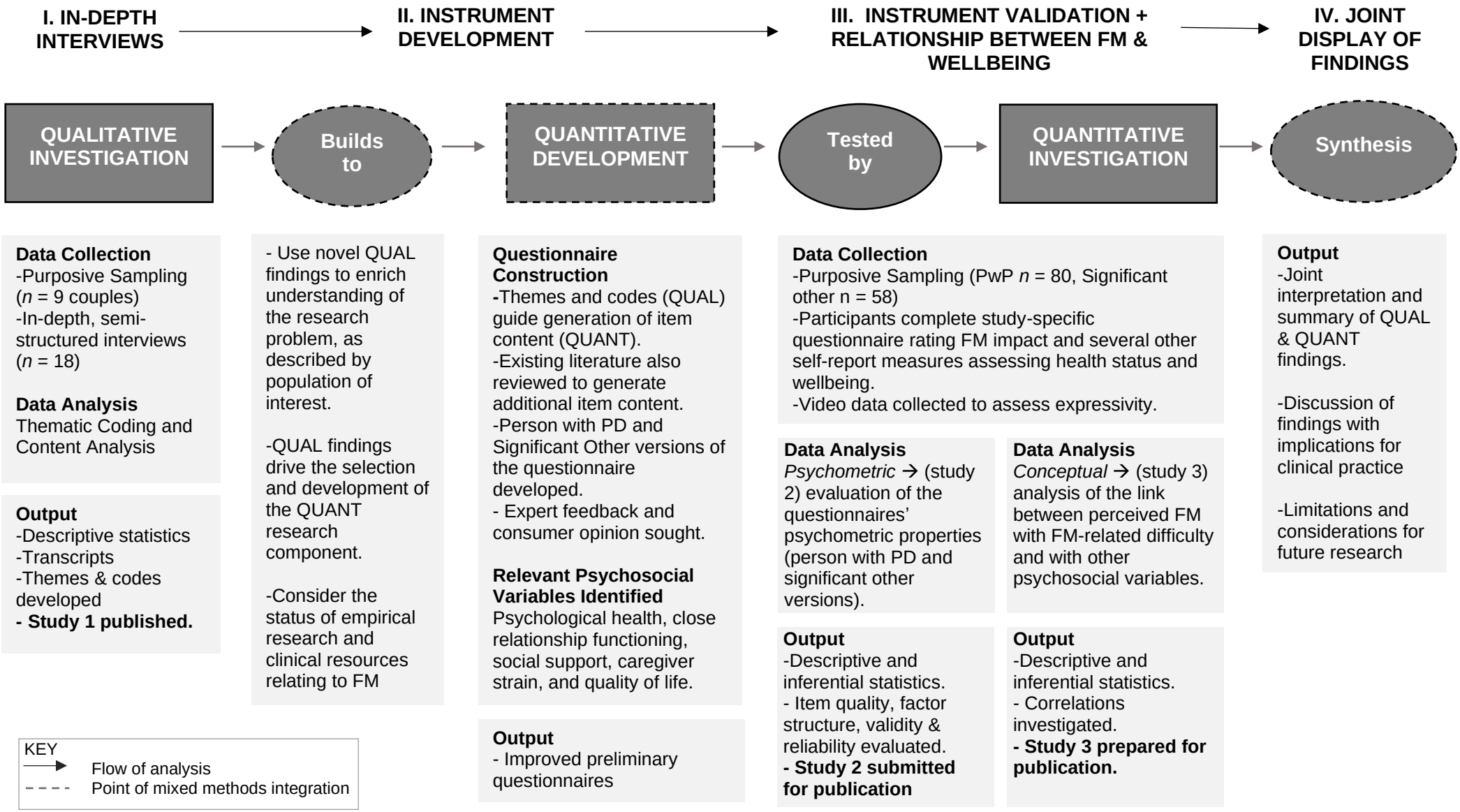
This sequence reflects an exploratory sequential mixed-methods design. The timing of the design is consecutive, meaning the quantitative phase was conducted after the qualitative data collection and analysis was completed. The research began with in-depth semi-structured interviews to explore individual accounts of facial masking and enrich understandings of expressive dysfunction in the context of daily life. It was important the initial step was exploratory since there was limited knowledge or theory to build upon at that time.

The subsequent quantitative phases of the research were then developed. The choice to construct a psychometrically valid clinical tool and the identification of relevant variables for further investigation, was driven, in part, by the qualitative findings. The quantitative phase can be further divided into psychometric and conceptually oriented studies. In the former, the qualitative findings were used to inform the development of two targeted self-report questionnaires (one for people with Parkinson's and one for significant others), followed by an evaluation of their psychometric properties. The latter investigation examined the association of perceived FM with psychosocial outcomes in PD. A summary of the studies included in the qualitative and quantitative phases is provided in the following sections, with each study described in further detail in their respective chapters of this thesis (4–6). The juncture between the qualitative and quantitative components of this research is the first point of integration, as the qualitative findings (themes and codes) were used to build the quantitative instrument (generate items for the questionnaires). The second point of integration occurs at the end of the thesis, where an interpretation of the qualitative and

quantitative findings is presented together (Chapter 7). Additional analyses which could not be incorporated into the separate papers for Studies 1-3 are also included in their respective chapters (4-6).

Figure 1

Diagram summarising key elements of the research design and methods, with interconnection to the thesis format.



Note. Figure adapted from Berman, E. A. (2017). An exploratory sequential mixed method approach to understanding researchers' data management practices at UVM: Integrated findings to develop research data services. *Journal of eScience Librarianship* 6(1): e1104, pg6.

Research Rationale & Positionality

A mixed-methods design was selected as it was considered best to accomplish the aims of the study, and because it aligns with the adoption of pragmatist and applied orientations. Pragmatism is an action-oriented process, which mixes research methods and presents a joint interpretation of research findings (Teddlie & Tashakkori, 2012). Taking up this orientation can be traced to my background in population health, clinical psychology, and more broadly to applied fields of social science, which emphasise practical application in the treatment of ill-health and the promotion of wellbeing.

To summarise, the basic assumptions of mixed methods are as follows: the nature of reality is both singular and multiple; a practical approach is taken to focus on addressing the research problem and the best methods to answer it effectively (Teddlie & Tashakkori, 2012). The process of research mixes inductive data (using personal perspectives to build common patterns and interpretations) and deductive data (testing a priori theory) (Creswell & Plano Clark, 2017). Mixed methods is distinguished by a cyclical method involving an exploratory process, where theory or variables may be generated from the data, and a confirmatory process, where theories are tested (Johnson & Gray, 2010; Denzin & Lincoln, 2011). By incorporating both types of findings, a fuller and more balanced account of a phenomenon can be provided (Morse & Niehaus, 1994; Onwuegbuzie & Leech, 2004; Onwuegbuzie et al., 2007; Creswell & Plano Clark, 2017). As recommended by Onwuegbuzie and Leech (2005), the framework of Greene, Caracelli, and Graham (1989) was used in defining the purpose of the core design: *expansion* (multiple standpoints or realities provide an enriched account of the under-researched phenomenon of FM than either method alone might provide), *development* (to use the results of the qualitative exploration of FM experiences to inform the construction of an instrument or intervention), and *complementarity* (to verify and elaborate on the results from the qualitative phase using the results from the quantitative phase).

There were three main reasons for the selection of a mixed methods design. Firstly,

the design is considered apt when little is known about the phenomenon or the variables of interest are largely unknown (Creswell & Plano Clark,2017), as is the case with the present research topic. Secondly, the selection of an exploratory sequential sequence was a response to the status of FM research at the time of planning this thesis. Beginning the research by gathering individual accounts of masking was important because the investigations of FM had been largely constrained to the social judgements of unfamiliar perceivers, rather than understanding the everyday significance of having this symptom or having a loved one with this symptom. In addition, giving voice to the perspectives and experiences of people who have FM was thought appropriate within the context of optimal PD management, which encompasses an equal consideration of medical treatment and the impact of living with the condition, both for people with PD and those they share their lives with (van der Eijk et al., 2011; Lim et al., 2017; Bloem & Brundin, 2020).

Thirdly, the decision to use an exploratory sequential design was also a response to the status of FM within PD care in New Zealand at the time of planning this thesis. Informal discussions with healthcare practitioners and PD support organisations at that time (around 2014) suggested FM was a topic given little clinical attention compared to other aspects of the disease. While the practitioners commonly observed FM, most had rarely spoken with their patients/clients about this topic. Rating tools available at the time (Goetz et al., 2008; Tickle Degnen, 2010) also did not provide evaluation of the impact of FM in PD. It was these observations, along with a pragmatist orientation, which contributed to the selection of an emergent design that could explore personal experiences of FM and apply relevant insights to the construction of a clinical tool sensitive to the consequences of this aspect of PD in everyday life.

Qualitative Phase Summary: Study 1

Study 1 aimed to explore personal accounts of people with PD and their partner's or spouse’s experiences of FM. In-depth, semi-structured interviews with people who reported FM and their spouse or partner were conducted. Study 1 strove to understand how this

symptom might influence psychological and social wellbeing of people with PD and their spouses and considered how wellbeing may be enhanced and managed. An approach of interpretive description was adopted to generate new knowledge that has clinical relevance and could make a difference to people who experience masking and their families (Thorne et al., 1997; Thorne et al., 2004).

Specific hypotheses were not identified prior to data collection, but assumptions about what types of topics might be important to people with FM and their families were derived from existing literature relating to facial masking in PD and other populations. Interview topics included the following: general/history information, personal impact of having FM, perceived impact of FM for their close relationships, reactions to FM in other social contexts, and ways of coping with FM.

Interview participants (*n* = 9 couples) came from a general community sample recruited through advertisements placed in national publications and a public hospital intranet in the North Island of New Zealand. Inclusion criteria included a diagnosis of idiopathic PD with masking status clarified when potential participants first made contact by checking whether reported descriptions of facial appearance were broadly consistent with markers of FM in PD. Face-to-face interviews were conducted separately with PD participants and their spouse or partner to allow for open discussion of potentially sensitive topics (*n* = 18 interviews). Telephone interviews were also conducted with subset of participants located outside of the central North Island of New Zealand (*n* = 12 face to face interviews, *n* = 6 telephone interviews). Analysis of the interview data followed the guidelines of thematic content analysis, with an inductive (or descriptive) approach employed (Braun & Clarke, 2006). A detailed description of the study is provided in Chapter 4.

Quantitative Phase Summary: Study 2 and Study 3

The quantitative phase of the research involved two studies, which shared the data collection phase, but were analysed separately and had different aims. Study 2 was psychometrically oriented, involving the development and validation of two novel self-report

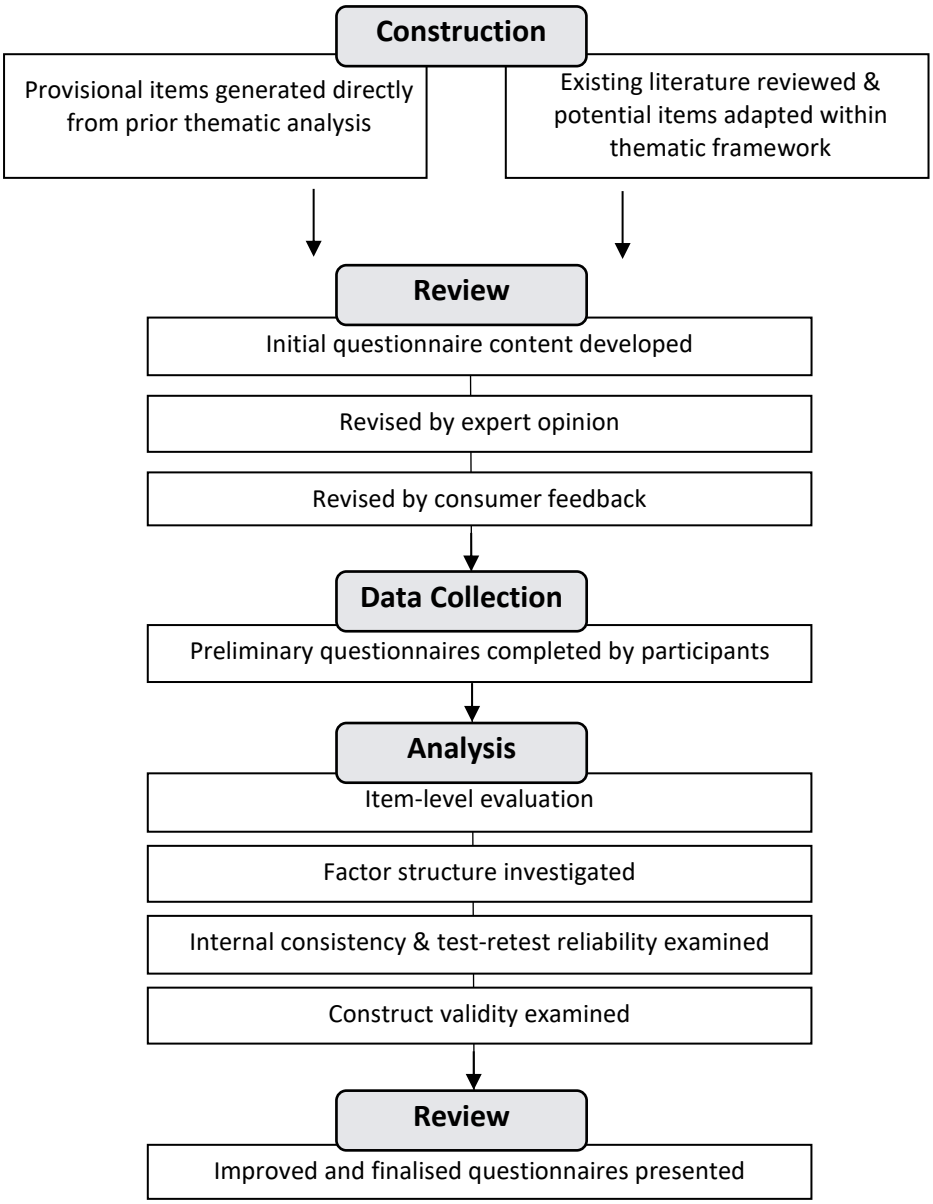
questionnaires to assess FM-related difficulty in people with PD and their significant others. Study 3 was conceptual, exploring the nature and strength of association between perceived FM with health and wellbeing outcomes in PD. In the (shared) data collection phase, a community sample of people who have Parkinson's (PwP) and their significant other (SO) were recruited from multiple centres across the North Island of New Zealand, via advertisements to PD support group members (*n* = 80 PwP, *n* = 58 SO). SO could include a spouse or partner, close friends, or family members (aged over 18 years). Eligibility was not based on FM status so that participants had a range of FM severity from normal expression to severe FM. Participants were asked to complete self-report rating scales assessing various aspects of health and wellbeing. PwP were able to optionally consent to a videotaped conversation with the researcher to assess the level of facial expressivity.

A flowchart breaking down the basic steps of Study 2 is provided in Figure 2, with a detailed description of the study materials and methods provided in Chapter 5. The aim of developing two new questionnaires was to aid understanding of the consequences of FM for people who have PD and their loved ones, and to provide a structured way to identify people that need help or support with this symptom. Thus, the first objective of Study 2 was to generate items for the preliminary questionnaires and evaluate the content validity, general feasibility, and clinical applicability. Item content was primarily generated from the interview data obtained in Study 1. Existing assessment measures were also reviewed for conceptually relevant content which could be adapted to fit the experiences of interviewees. Two separate forms of the questionnaire were developed: one for people with PD, and one for significant others. The pre-test version of the two questionnaires were revised through expert consultation and feedback from people with PD and their partners from the original interview sample. The second objective of Study 2 was to statistically evaluate the psychometric properties of the two questionnaires. The revised preliminary questionnaires were then administered to a sample of people with PD and their significant others (participants who had PD *n* = 80, significant other participants *n* = 58) on two separate occasions around 4–6 weeks apart. Psychometric testing of the questionnaires included an

item-level evaluation, factor structure, reliability, and validity testing. A final version of the questionnaire was then developed based on the psychometric findings and suggestions for future revisions compiled. Supplemental data, which was submitted for publication but could not be included in the main manuscript for reasons of brevity, is presented in separate appendices for Study 2.

Figure 2

Flowchart summarising the procedure of Study 2



There were two broad aims of Study 3. First, to explore how FM is perceived by people with PD themselves and their family members. Four groups provided ratings of FM severity: PwP and SO who separately reported facial masking severity on a study specific self-report measure; Parkinson's nurses who provided clinical assessment of FM staging from a videotaped conversation; and a researcher-rated evaluation of facial expressive behaviour using the videotaped conversation. The second aim was to examine the effects of FM on wellbeing, by examining the association between FM severity and various psychosocial outcomes for PwP and SO. Other factors which impact psychosocial outcomes in PD were also examined. Self-report questionnaires measuring psychological health, social functioning, and quality of life were completed by people who had Parkinson's and their significant others. Supplementary data on the coding procedure for the video is presented following the manuscript.

4. Study 1
Unmoving and Unmoved, Experiences and Consequences of impaired non-verbal expressivity in Parkinson's patients and their spouses

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ORIGINAL ARTICLE

Unmoving and unmoved: experiences and consequences of impaired non-verbal expressivity in Parkinson's patients and their spouses

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ABSTRACT

Purpose: Hypomimia, or facial masking, is a decrease in voluntary control and spontaneous movement of the muscles of the face, which may occur in Parkinson's disease. Little is known about the psychosocial consequences or management of this symptom. The aim of this study was to provide an initial overview of patient & spousal experiences of living with an acquired nonverbal expressive impairment in Parkinson's disease.

Method: This qualitative study involved a community sample of individuals with Parkinson's who experienced facial masking, and their close romantic partners. Nine people who had Parkinson's and nine of their spouses or partners participated in separate (individual) semi-structured interviews.

Results: A descriptive thematic approach was used to analyze the interview data. Key themes relevant to rehabilitation included the misidentification of masking as negative affect, poor symptom recognition, and unmet health resource needs.

Conclusions: The results indicate masking can have an adverse impact on close relationships and psychological well-being. This study presents an initial basis for clinicians working with Parkinson's populations to recognize of the needs of people who experience masking and better support such individuals and their families to live satisfying social and emotional lives.

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KEYWORDS
Hypomimia; facial masking; facial affect; emotional expressivity; psychological wellbeing; psychosocial management

➤ IMPLICATIONS FOR REHABILITATION

- People who have Parkinson's and their families are often unaware facial masking is a symptom of Parkinson's disease.
- Masking may be confused with negative affect and become a barrier to satisfying close relationships.
- Masking related health resources and support are perceived to be of poor availability and quality.
- This qualitative study suggests masking be considered as an etiology of interpersonal and psychological difficulties in Parkinson's disease, and encourages better recognition of this unique population's health education needs.

Introduction

Parkinson's disease (PD) has recently been identified as the fastest growing neurological disorder, exceeding the growth of conditions like Alzheimer's disease [1]. In New Zealand, population structures are ageing [2] and because PD is a mostly a condition of later-life [3], the number of New Zealanders who have PD is expected to rise. This growth creates a need to extend the duration and scope of PD care.

Hypomimia, or facial masking, is characterized by the loss or reduction of facial expressivity and is a distinctive feature of PD. The loss of facial expressivity is caused by an increased rigidity and slowness of muscular movement in PD, which lessens the capacity to convey emotions, thoughts and characteristic behaviors [4]. Scientific literature has largely ignored patients' perspectives in investigating facial masking. It is this gap that likely contributes to the scarcity of research on the psychological and social significance of this symptom, and lack of evidence regarding the effectiveness of rehabilitation approaches.

The prevalence of masking within PD has not been well researched but is estimated to range between 39% and 65% of patients, depending on age and disease progression [5]. The subjective experience of emotion is thought to remain intact in PD [6], but deficits in emotion recognition & processing have been reported. Impaired facial expressivity in PD may be in part associated with these deficits [7].

The role of facial expression in communication has been well-researched in the scientific literature [8]. In close relationships, nonverbal facial expressions, particularly displays of positive emotion, are crucial to relational processes such as conflict management and maintenance of intimacy [9,10]. Research has explored the social significance of facial paralysis. For example, in Moebius syndrome, where there is congenital loss of facial motor movement preventing the display of emotion and other communicative gestures [11–13]. The loss of facial expressivity may be especially significant in PD, because there is often simultaneous impairment of other channels of communication, such as speech and bodily gesture.

Research on the communication of people who have PD has suggested that the reduction of facial expressivity could cause individuals to be misjudged, appearing cold, moody, unfeeling, avoidant or even intellectually impaired [14–16]. The loss of facial expressivity can have an adverse influence on impressions of

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people who have PD. For example, naïve older adult observers (strangers) who viewed short video clips of people with PD were less interested in relationships with individuals who had higher masking than lower masking, and perceived those with higher masking as less socially positive [16]. Student health professionals who viewed silent video clips were also found to make more negative judgments of affect, personality and intellect of people who had PD than individuals with ischemic heart disease [14]. Experienced health professionals have been found to evaluate individuals with a higher degree of masking as less cognitively competent, less supportive and less sociable than those with lower masking [17], and form more negative impressions of some personality traits in those individuals who experienced masking [18]. These findings suggest a significant impact of masking in the context of the health care, yet little is known about the consequences of masking for people with PD or within close/familial relationships. A single previous study found that masking was negatively associated with the social well-being of care partners, and with the quality of the partner's relationship with the individual who has PD [19].

If healthcare professionals are susceptible to forming inaccurate impressions of people with masking, spouses may also have difficulty accurately detecting what their loved one is thinking and feeling. On the other hand, a heightened familiarity between individuals in close relationships may mitigate the effect of masking. This is the subject of the present study, which offers an initial exploration of impaired nonverbal expressivity from the perspective of people who have PD and their spouse or partner. More specifically, this study strove to understand how this symptom might influence psychological and social wellbeing of people with PD and their spouses. The study also considers how that wellbeing may be enhanced and managed. Therefore, the study takes an approach of Interpretive Description, seeking to address a "practically relevant knowledge gap" [20]. This study design enables a description of the phenomenon of interest, but also aims to generate new knowledge that has clinical relevance and can make difference to people who experience masking and their families [21]. Psychological and social wellbeing are pertinent topics in PD management because of the chronicity of the condition and long duration of care usually provided by family members within the home. The progressive and debilitating nature of the condition increasingly restricts participation across employment, sporting, leisure, and social domains. As the disease advances, family and home life may become the focal point of activity, meaning and purpose. Masking has potential to jeopardize communication and meaningful social connection, and in turn, patient and carer quality of life. Addressing this symptom is, therefore, a potentially valuable endeavor in PD management. The objectives of the present study called for an experiential qualitative approach, which identified salient and recurring patterns or themes across individual experiences. Semi-structured interviews were chosen for their ability to provide a detailed view of people's everyday worlds [22] and for their encouragement of participants to speak openly about a particular aspect of their lives [23].

Method

Ethical approval was received from the University of Waikato, School of Psychology Research and Ethics Committee, prior to commencement. The study formed part of a program of doctoral research utilizing a mixed method (exploratory sequential) design, with a main theoretical approach of critical realism. The lead researcher's joint background in Psychology and Population Health framed the wider project, which focuses on practical clinical applications with an aim to improve patient outcomes and clinician recognition.

Participants

Participants were people with PD who experienced facial masking and their romantic partners ($n = 9$ couples), who came from a general community sample in the North Island of New Zealand. Advertisements seeking people with PD (and their partners) who experienced changes to facial expression, appearance, and movement were placed in Parkinson's community support group newsletters, a health magazine, a hospital staff webpage, and public hospital reception. Local news media publications also generated interest.

Individuals or their partner/spouse were required to have been diagnosed with idiopathic PD by a healthcare professional, usually a Neurologist. Masking was self-reported by participants. Masking status was further clarified during eligibility screening where potential participants were asked if their facial appearance was broadly consistent with markers of masking in PD (ascertained by the first author when potential participants first made contact). These markers were derived from the Unified Parkinson's Disease Rating Scale [24] and the Interpersonal Communication Rating Protocol: Parkinson's Disease Version [25]. People who had PD often reported uncertainty or poor awareness of their own facial expressivity, thus partner accounts of facial appearance and movement were prioritized in establishing eligibility. Exclusion criteria were a diagnosis of dementia or any other impairment that would prevent comfortable participation in an interview, and any non-PD-related alteration in facial appearance. Participants were required to have been in a close romantic relationship of 6 months or more. Eleven couples were deemed eligible for the study, but two of these couples opted out for personal or medical reasons. In New Zealand, treatment of chronic conditions like PD is publicly funded and many aspects of care are free or subsidized. Most participants had seen a Neurologist. Typically, participants' managing doctor was a General Practitioner. Several participants were active members of their local Parkinson's Society, which is primarily staffed by Registered Nurses in New Zealand.

Data collection

Semi-structured interviews were conducted individually with each member of the couple, to allow for open discussion of sensitive relationship issues. Participants were asked to choose a private location and interview time that suited their medication schedule and any preferences related to their condition among other things. At initial contact, potential participants were provided with verbal information to increase the likelihood they understood the nature of the study and were screened for eligibility. Eligible participants were then provided full written information about study participation and given an opportunity to ask questions about participating. If appropriate, an interview time was set up. At the interview appointment, key information about the study was verbally reiterated and any participant questions answered fully before consent was obtained. The interview topics included general/history information, experiences of facial masking, individual/relational impact, and coping with masking. Topics were introduced by open-ended questions, which were followed by probes to encourage elaboration, clarify meaning, or further investigate a particular topic. To minimize distress and provide balance to the emotional tone of the appointment, interviews concluded with discussion of pleasant aspects of interviewee's lives. A list of local agencies who could provide support was supplied following the

Table 1. Participant characteristics.

Participant code	Gender	Participant with PD or partner	Age group (years)	Age at PD diagnosis (years)	Expressive impairment duration (years)	Interview type (in person or telephone)
P01	M	PDparticipant	50–59	50–59	2	IP
P02	M	PDparticipant	60–69	50–59	4	IP
P03	M	PDparticipant	40–49	30–39	5.5	IP
P04	M	PDparticipant	50–59	40–49	8.5	T
P05	F	PDparticipant	60–69	50–59	1.5	T
P06	M	PDparticipant	70–79	60–69	3	IP
P07	M	PDparticipant	60–69	60–69	7	IP
P08	M	PDparticipant	60–69	60–69	5	IP
P09	M	PDparticipant	60–69	50–59	5.5	T
P10	F	Partner	60–69	–	–	IP
P11	M	Partner	60–69	–	–	T
P12	F	Partner	30–39	–	–	IP
P13	F	Partner	50–59	–	–	T
P14	F	Partner	60–69	–	–	IP
P15	F	Partner	60–69	–	–	IP
P16	F	Partner	50–59	–	–	IP
P17	F	Partner	50–59	–	–	IP
P18	F	Partner	60–69	–	–	T

interview. Telephone interviews [6] were conducted with participants located outside of local regions. These followed a similar procedure, with eligibility and consent completed verbally over the telephone. A copy of the completed verbal consent form was supplied following telephone interviews. Interviews were generally well tolerated, with four people with PD opting to take short break. Interview length ranged from 38 to 105 min, with most around an hour in duration. Telephone interviews were on average 14 min shorter in duration than face-to face-interviews. Interviews were audio recorded and transcribed verbatim, reproducing all spoken words and sounds. Transcript extracts presented here are lightly edited to improve brevity and readability, by removing non-essential words or phrases, without altering meaning. Editing is indicated by three full stops in a row. The analysis was carried out with the full, unedited transcripts.

Data analysis

Interview data were analyzed using thematic content analysis, following guidelines set out by Braun and Clarke [26]. An inductive approach to thematic data analysis was employed. This approach is predominantly descriptive and involved analyzing the data without a predetermined structure or framework which could bias the analysis and limit theme development [26]. Given the paucity of scientific literature about this phenomenon in the context of patient/spousal populations, this approach was considered appropriate. As earlier studies have been quantitative and concentrated on health professional or naïve observer populations, this study aims to give voice to patient and spousal experiences of masking.

Data analysis was carried out by the first author of this paper. This followed Braun and Clarke's [26] six phase procedure: (i) *Transcription and familiarization with the data.* Verbal data were transcribed by the first author and read to familiarize the researcher with the data. Where audio recordings were unclear, the second and third authors provided consensus. (ii) *Generating the Initial Codes.* Data extracts were systemically coded and similar codes collated. Data were initially coded in an open, descriptive approach before a more focused reorganization and re-categorization was performed. Coding emphasized participant voices, often using phrases or words derived from the data. We deviated slightly from Braun and Clarke's approach to take account of the saliency of codes. Consistent with the aims of interpretive description, this process addresses individual variation and patterns of thematic commonality, by allowing important but non-recurring themes to be taken into account [21]. This approach produces an understanding of the phenomenon that describes the complexities which occur real-world health settings, in a way that may assist clinical practice [27]. (iii) *Developing the Themes.* In this stage, codes were sorted into possible themes and all relevant coded extracts collated within potential themes. Codes were examined to investigate whether they form a coherent overarching theme. Themes were developed based on analysis of all interviews conducted. Initial codes and themes were discussed with the second and third authors of this paper to ensure coherence. (iv) *Refining the Themes.* Each theme and collated data extracts were then reviewed for consistency. Themes were refined, revised or discarded. Thematic relationships and coherency of themes within the entire data set was considered, to ensure the analysis accurately reflected the whole data set. (v) *Defining the Themes.* Theme names and definitions were examined to check what aspect of the data they reflected and were reworked and refined. (vi) *Narrative Development.* In the final stage, an analytic and interpretative narrative was developed which described and related together the themes and subthemes, using illustrative data extracts. Thematic and narrative development was reviewed by the second and third authors and further refined by the first author. Interviewees were invited to comment on initial interpretations of the general interview data, to ensure nothing important was left out or overemphasized and to provide opportunity for clarification. No additional comments were offered.

Results

Participant characteristics

A description of the participant cohort is provided in Table 1. All participants who had PD and experienced facial masking (hereafter "PDparticipants") were taking antiparkinsonian medication at the time of study participation. One PDparticipant had received deep brain stimulation approximately 1–2 years prior to study participation. At the time of interviewing, most were generally independent in activities of daily living.

Most PDparticipants were male. Four PDparticipants had been diagnosed before age 55 years. Most PDparticipants had a disease duration (time since Parkinson's diagnosis) of less than 10 years. The length of time with facial masking ranged from 2 to 9 years. Three PDparticipants were employed at the time of interviewing. Most couples were married and living together in private homes. Relationship length ranged from 4 to 45 years. Two couples

Table 2. Thematic analysis outline.

Theme	Subthemes
1. Unmoving and Unmoved: Disturbances of Facial Movement and Social-emotional Expressivity	○ *"Watered down"*: Muted and Distorted Expressions ○ The Discomfort of Facial Difference: The Absence of Expression ○ Inauthentic and Effortful: Voluntary Expressions ○ *'I can see him, but I can't'*: Facelessness
2. *'If that information's not there, you fill it in'*: Misinterpretations of Negative Affect	○ Misattribution of Negative Affect
3. *'Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection	○ *You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity.
4. *'It can affect you quite badly without you realising exactly what it is'*: Poor Symptom Recognition and Understanding	○ *'They don't get that information because it's not there and because the harm that's already done in a relationship isn't understood at all'*: Unmet Health Resource Needs
5. *'Almost an Asset'*: Positive Reappraisals of Masking	

established their relationships after or around the time of PD onset. One couple was raising young children and another young adult children. There was little variation in the ethnicity of the sample cohort, with all participants reporting themselves of New Zealand or Australian European descent. Major themes from the analysis are summarized in Table 2.

Unmoving and unmoved: disturbances of facial movement and social-emotional expressivity

This theme describes how the interview participants ("partners" and "PDparticipants") conceptualized disturbances to facial appearance and movement, which could impede communication. On a deeper level, it illustrates how such disturbances obscured the expression and identity of the person with PD and could obstruct emotional comprehension and social connection. Several subthemes were identified:

"Watered down": muted and distorted expressions

Participants described a diminishment of the intensity of emotional displays or a reduction in the size of facial movements. They also described a slowing in execution, and delay in initiation, of facial expression or movement. Muted and slowed expressions could make partners feel cut off and confused. This seemed to occur when a discrepancy between nonverbal and verbal communication led to a difficulty accurately comprehending affective state and/or communicative intent: *"It's delayed, even when it's spontaneous… if he's not happy, he seems to have lost that… You know not a gradual change, it's more a delay but then quickly goes into it. You can't really judge when you're talking or something like that, where you are" (P17 – Partner).*

Abnormalities in the appearance of positive affect displays, especially a smile, were repeatedly highlighted. Voluntary smiling, and to a lesser extent spontaneous smiling, were described as distorted or grimace-like. These expressions lacked in emotional intensity and characteristic muscular movements, such as the involvement of eye muscles. For example: *"he will do something that looks a bit like a smile. He can probably move his lips up… it never kinda goes to his eyes anymore. You know when you say someone smiles with their whole face, with their eyes… A smile to the eyes is a really unusual, it's quite a special thing now" (P12 – Partner).* Spontaneous natural smiling was reported to become increasingly infrequent and often spoken of somewhat wistfully by partners of people with PD, highlighting the value they placed on positive expressions.

The discomfort of facial difference: the absence of expression

A more global absence of facial expressivity was also reported by most participants. An unchanging or immobile facial appearance could be perceived as blank or emotionless. Because their underlying affective state or intent was concealed, this appearance led partners to think the person with PD was unmoved or unfeeling. In actuality, people with PD may experience strong affect and partners may feel disconcerted at the incongruence of verbal and nonverbal communication: *"It's interesting, he can be really upset and your face wouldn't tell anything. You know it's that real sort of, dead pan face. You know, he can be saying the words but the face doesn't tell, it doesn't tell anything" (P13 – Partner).* A sense of unease and social discomfort was often associated with this blank appearance for both observers and PDparticipants. The blank appearance was also commonly accompanied by an impression the individual was staring: *"it's not only a mask, it's sort of a blank stare, and sort of like having someone staring at you can be quite uncomfortable" (P17 – Partner).* This seemed to occur when a reduced blink frequency was combined with an absence of facial expressivity, causing partners (and other individuals) to feel uncomfortable or intimidated when interacting with the person with PD.

PDparticipants also reported that their unusual facial appearance sometimes led to unpleasant social interactions with colleagues or strangers. Observers' reactions could be unkind and even humiliating, as they misinterpreted the cause and meaning of reduced social expressivity. One PDparticipant recalled being taunted for the way he looked by a group of adolescents on public transport, another described being confronted at work by a colleague: *"I can remember him saying several times in the middle of meetings you know, 'I can't read you, your face is blank. You never smile'… He said, 'your face would crack if you even tried to smile!'" (P07 – PDparticipant).* Embarrassment or discomfort at having a facial difference also influenced PDparticipants on an intrapersonal level. The same participant (as above) later described his concerns of how his facial appearance might be evaluated by others, which could lead him to withdraw: *"when you're in a, sort of a group of people, ah that you're not friends with or know well, and you sometimes feel that they're looking at ya as though to say, 'God he's a gormless wonder now!' So, (laughs), that's what I think they're thinking and that alters what I feel, which means I'll drift to the back of whatever's happening".* This participant did not mention being humiliated by others for his facial appearance in a social context but had previously experienced colleagues mocking his gait and tremor. Having a facial difference may further accentuate the difficulties of living with a chronic illness and physical disability. Unpleasant interpersonal interactions with individuals who misinterpret reduced expressivity, or do not recognize it as a symptom of the condition, can increase the social isolation of people who have PD from their families and communities.

Inauthentic and effortful: voluntary expressions

One method participants used to compensate for diminished (spontaneous) expressivity was deliberately posing expressions;

however, this could be experienced by PDparticipants as unnatural or forced. The process of posing an expression was described as mentally and physically exerting. The effort of presenting a smile was particularly emphasized, *"it's a matter of trying to coordinate a whole lot of muscles… So to smile, you're having to go through a whole thought process as to how am I going to do it. Because it doesn't come naturally" (P06 – PDparticipant).* The effort of voluntarily posing socially appropriate expressions prevented many PDparticipants from using this strategy regularly. Voluntarily posed expressions were also experienced as false. This feeling was most often associated with the effort required to produce a movement once natural and automatic, or with the sensation that a posed expression was forced and artificial. For example, *"it should be happening normally, it doesn't happen normally… feels false. The nature of the smile is uh, not the time you're actually putting in, not how much effort you're putting to it" (P04 – PDparticipant).* The perception of artificiality could also prevent PDparticipants from using this compensatory behavior. Interestingly, the only female participant with PD reported no concerns about her expression seeming false. She commented that posing facial expressions was effortful but preferable to appearing angry or sad, and points out her motivation for doing so:

> *I think I try and, and keep it a positive (pause) outcome (pause) you know I'm sort of very conscious of the fact that we spend a lot of time together now that we're sort of retired….If I put myself in his shoes I couldn't bear to you know sort of come in and find sort of a sad person sitting there with a grumpy face, for whatever reason… it would um, sort of almost unthinkable outcome to be constantly dreary and complaining and that… with the facial masking… I think as long as I've still got the ability to try and um, influence it, I would do that, rather than just let it sort of, compound, as a grumpy face. (P05 – PDparticipant)*

"I can see him, but I can't": Facelessness

Partners did not explicitly report observing posed expressions as inauthentic or effortful. Partners' perceptions focused on the difficulty 'seeing' their partner through muted, distorted expressions or a blank facial appearance. Their responses were typified by feelings of loss, unfamiliarity, and uncertainty. The impairment of expression is far more than a reduction of facial mobility for these couples. Facial expressivity facilitates communication, but faces are also a key means to displaying self-identity and sustaining connections with others: *"I've lost… some aspects of that person-… the face is definitely one of them. He's still got a lovely face, but… I can't read that face anymore… I can see him, but I can't… It's definitely still him, it's still his lovely face… I just have to really look to see what that face is telling me, you know" (P13 – Partner).*

For PDparticipants, this facelessness was represented by an incongruence between their blank outward appearance and their internal emotional experience, often first observed when viewing recent family photographs: *"I don't know who it is. It's not me…. I used to be able to express myself in face and laugh and things like that, I don't know. I look at photographs of me, and I don't think they're me at all… I don't believe it's me" (P09 – PDparticipant).* This feeling was deepened when observers seemed to overlook PDparticipants, possibly because of the lack of expressivity. The loss of identity and self-expression is particularly important because PD restricts physical participation in those activities which individuals define themselves by, such as sports/leisure activities and employment. PD neutralizes the identity and vitality people with PD present to the world: *"You don't see me as exceptional person or as a person not exceptional… Because I used to feel … somewhat exceptional… Like I did [extreme sports] and ah, it was always associated with some sort of verve…. I don't really feel like I'm trying to achieve anything, no one raises any comments or shows any indication that I'm plus or minus" (P04 – PDparticipant).* A sense of becoming unremarkable and unnoticed was echoed by partners. The perception that these consequences of masking could not be addressed was common, highlighting the helplessness and sorrow that may be felt by both families and people who have PD:

> *He said that… 'I often feel that people don't see me'… even though he hasn't changed… his voice has changed… you don't notice his face so much, you know. His face kinda blends into the crowd if that makes sense. I think that has had a big impact too and… it's something he can't change… there's nothing he can really do to make that any better. You know, which is the hard thing really. (P13 – Partner)*

'If that information's not there, you fill it in': misinterpretations of negative affect

When there were no verbal cues to interpret, partners confused a lack of facial expression with negative affect: *"if that information's not there, you fill it in and everything you fill in no facial expression with is boredom, tiredness, anger. The really negative emotions" (P12 – Partner).* Partners most commonly misidentified reduced expressivity as anger, lack of interest, boredom, disapproval and sadness. The misidentification of negative affect also occurred in other relationships, e.g., colleagues, friends and children or grandchildren. Around half of the partners in the study reported they misinterpreted masking in this way. Those who did not, tended to describe their loved one's facial appearance as blank or unemotional (see previous theme). PDparticipants also clearly reported that their facial appearance did not always reflect their internal experience, *"I just know that I feel good inside and… I want to express myself out, but my face isn't expressing anything" (P09 – PDparticipant).* In other words, the impassive or negative affective states identified by partners resulted from misinterpreting an unmoving facial appearance to have communicative or emotional meaning. Interestingly, nearly all partners seemed to realize their identifications of negative affect were incorrect. Despite this, misidentifications of negative affect were persistent. For example, one partner describes having to backtrack and remind herself her perception of her husband's mood is likely incorrect: *"I've had to kind of pick myself up because I've looked at [him] and thought, oh my God, he's not interested at all! But then I remember he probably is, but… it doesn't show… I said to him one day, he looks like he's bored sh*tless! And, and he's not, but that's how the face can sometimes look. You know, and he's not, not really" (P13 – Partner).*

Misattribution of negative affect. Some partners not only had difficulty accurately identifying PDparticipants' emotion, but also reported a tendency to form incorrect attributions about the cause of negative affect they observed. The nature and content of these attributions varied, but partners usually ascribed perceived affect to failings in their own behavior, or to a decrease in the person with PD's relationship interest/satisfaction. Partners' reported this cognitive process persisted despite knowledge the PDparticipants' affective state was likely concealed by masking. For example, a few partners described persistent thoughts the apparent negative mood was their fault, or signaled a personal criticism or rejection:

> *Sometimes he was looking disapproving … I was being a bit apologetic. And he said to me one day, 'why do you keep apologizing? I can't understand why you keep apologizing to me all of the time'. And I said, 'because I think you're upset with me in some way'. And he said, 'but I'm not!' (Laughter)… Also we've had an awful lot of adjustment relationship-wise… we have had a lot of decisions to make and um, I think it's very easy to make assumptions in a relationship that… might be disapproving*

in some way which they're actually not. … When I really think about it, and we talked about this since, that I've often made assumptions [he] was annoyed with me in some way when he wasn't … I'm very sensitive about disapproval because I had so much of it for so long. And I'd never had that with [him]. So I was confused I think, yeah. (P14 – Partner)

Not all partners in the study misidentified masking as negative affect and misattributed its meaning. Those who did also tended to report a higher degree of expressive impairment (i.e., minimal spontaneous or voluntary facial movement and/or a fixed expression much of the time). A few partners also reported difficulty challenging the thoughts they experienced in response to masking, which they could find both distressing and dysfunctional. For example, this partner described her ongoing struggle with misidentifying negative mood and hints at how she attributes this to failures of her own behavior: *"I have to remind myself all the time that what I think he's feeling is probably not what he's feeling. I struggle with that personally, just every day, like sometimes I think 'oh God, this guy is just so miserable… God, I just can't seem to make this guy happy'. I have to remind myself that actually um, he's not, he's not unhappy because of me and the kids … Check in with him a lot … Remind myself of all the good things … There's no reason for him to be unhappy … just constantly reminding myself." (P12 – Partner).* This extract also illustrates the automatic and compelling nature of the cognitions experienced by some partners, in response to their misidentification of negative affect. Distressing and inaccurate judgments of facial behavior were also reported to occur prior recognizing masking as a symptom of PD. For example, a small number of partners feared the person with masking was considering leaving the relationship or was unfaithful.

'Your connections between people are being chipped away at': social-emotional distancing and disconnection

Mutual feelings of social or emotional distance and disconnection were described by just over half of participants, which they attributed in part or wholly to an expressive impairment and the difficulties associated with it. Generally, a face that appeared unmoving and inexpressive was thought to erode the meaningful connections couples shared. Often, the loss of expression was appraised as reflecting dissatisfaction with others' behavior, or a lack of care for the relationship. Prior to recognition of masking as symptom of PD, perceptions of detachment commonly resulted in withdrawal and frustration for partners and PDparticipants alike:

*I think that stuff like becomes self-fulfilling … with that diminishment in subtle signs of caring, I, I think (pause) self-protection kicks in … For her. And I think self- protection itself creates distance and it er, and it elicits response from me. Which is a bit, oh well then, what-the-f**k- ever, you know? … so distance develops … .it never became "oh f**k you I'm leaving" or anything like that, but it was, put it this way, it was probably, I would say it was probably a little bit of relief, for [her] to understand that some of that distance or, um wasn't being created by me, it was just, I wasn't being reflected to her. (P02 – PDparticipant)*

Masking did have an impact on how the above PDparticipant perceived distance developing in his important relationships, but his experience was also influenced by features such as depression and a lack of understanding of what was causing his difficulties: *"I don't want to overstate that, you know. But it was definitely there, but there was that along with feeling like sh*t um, and not knowing why, and all those things mixed up together, was hard, it was really hard actually … To the point of it … feeling you know in despair. Which is not my usual state … I was at risk there for a while" (P02 – PDparticipant).* By contrast for partners, prior to diagnosis of PD, masking could play a significant role in feelings of anger, shock and loneliness at what they observed was their loved one's rejection or disfavor of the relationship:

I would think did he not hear what I've said, does he not care that this is so exciting … he would think that he was being animated. So I was getting angry … And yeah, and so I stopped talking. I stopped telling him stuff … because I thought he was disapproving or I thought that he was disinterested … .Incredible loneliness for me, like a real sense of um, I don't like where this relationship is going … you know like whoa! This is not who we are, or what, how I communicate, or yeah. It was really quite painful. (P10 – Partner)

By the time of interviewing, most participants identified masking (or their responses to this symptom) as responsible for observed changes to relationship quality or the person with PD's behavior. While diagnosis and symptom insight alleviated the situation described by the individuals quoted above, other participants reported ongoing difficulties. A handful of participants used the metaphor of a barrier or wall, where impaired expressivity was likened to a feeling of being cut off from one's most important relationships. This was often characterized by a decrease in the readiness and ease of communication but could also have more significant implications. For example, this PDparticipant speaks of the difficulties of communicating when nonverbal cues are not visible, and the reciprocal isolation and avoidant behaviors which could result: *"Like we're in a dark room or whatever, cause she can't see anything she's got to listen to really what I'm saying … I find it very frustrating … there's something happening that's isn't being passed through by your face or whatever … It puts … a couple apart. It can … put [her] in a, in a place where she doesn't wanna be with me or doesn't wanna to talk to me and I'm in a version of the same. I can't talk to [her], or don't want to talk to [her]" (P04 – PDparticipant).* Other participants echoed this mutual disengagement and stressed the notion this process was difficult to quell, because the cognitions and behaviors driving it were somewhat automatic or habitual. The comment below illustrates one couple's mutual feelings of detachment and how this partner's perception her husband is unhappy or uncaring can easily snowball, if uncontested.

So if someone doesn't look happy when you walk into the room, or happy to be spending time with you, or animated while you're talking to them, very hard for me to feel emotionally connected to them. So I think it's had a huge impact … He would say that I'm distant, but then I'd say, (laughs) 'you're distant too' (laughs). I feel like there's this big block wall there sometimes and I've done everything I can and it's so, so much of it is subconscious. And it's not until I'm feeling really disenchanted, or disheartened, or really tired and upset … that I call myself back and go, but he hasn't said anything or done anything that would show that he was unhappy. (P12 – Partner)

Partners' comments tended to focus on how an unresponsive facial appearance could make people with PD seem uncompanionable and disengaged from the relationship: *"His expression doesn't change whether I'm in the room or not … that can be lonely. Just creates this barrier … this distance, it's like this gulf between us or before we've even started any conversation" (P12 – Partner).* Partners also perceived interactions to be less frequent in occurrence, which accentuated feelings of loneliness and being uncared for. A reduced capacity to share a satisfying and close companionship further isolated couples from one another, straining relationships through both physical and emotional distance. The following extract illustrates how deeply a masked facial appearance can divide and isolate couples.

INTERVIEWER: What's it like for you to live with [his] masking?

PARTNER: Oh it's miserable, it's miserable. Always with him, when you walk into a room and you get home from work and he doesn't seem to even notice that you've walked into the room (pause). It's quite hard to keep walking into a room, (laughs), when someone appears not that interested that you've walked into it … And I will, maybe not so much

avoiding interaction with him, but making sure I've got lots of interaction with other people. So I work full time really deliberately cause I can't be at home with him full time, and in fact, by the end of weekend I'm ready to come back to work and have that interaction again. (P12 – Partner)

It also highlights masking's influence on partners' mental well-being, and the implications for fulfillment of partners' emotional and social needs, in addition to being a carer. Many partners were at a life stage where they were retiring, spending more time at home, and found their roles as caregivers were increasing. The emotional distance created by masking was a further stressor to balancing the responsibilities of being a carer to someone with a chronic illness and being a romantic partner.

Not all participants provided such vivid descriptions, but it is pertinent to consider such comments within the context of PD. It is a chronic and progressive condition which limits physical agency, but also excludes sufferers from partaking in many domains of life. An impairment to facial expressivity has the potential to heighten isolation and exclusion from participation in an especially meaningful sphere of people's lives – their social relationships. The following extract provides a dramatic example of the importance of interpersonal wellbeing for people who have PD. On the flip side, it also illustrates this participant's lament for the unfairness of his family's suffering.

To have, you know, the physical symptoms and the masking and the whole blunted emotional affect … impact so heavily … on interpersonal relationships with my wife, with my kids … It's like a desperate fight to the end … The only thing I've got left is my family and my close friends … It's the only thing left in my life and it's what I'm trying to do … And to have that impacted by … an emotional sort of stonewall um, yeah it's really depressing. It's really hard … I just feel very short changed. I feel very short changed for my family, you know. Because … they didn't sign up for this type thing, you know. (P03 – PDparticipant)

'You haven't got someone who's excited when you're excited, or sad when you're sad': reduced emotional reciprocity

Partners reported a reduced reciprocation of supportive and rewarding interpersonal exchanges, *"It has I s'pose affected the relationship … You don't get that automatic feedback, and therefore you can't just go with the feel of the moment … We have had to bring in verbal checks … (spoken quietly, looking down) which sometimes takes away from the moment, you can't just go with the feel of it". (P17 – Partner).* This subtheme was observed in those partners who did not report confounding masking with negative affect. A reduced synchrony of social and emotional behaviors during interpersonal interactions contributed to partner feelings of loss and disconnection, and adversely impacted perceptions of their own general wellbeing. For example, *"It does impact on your enjoyment of life. Because you haven't got someone who's excited when you're excited or sad when you're sad … You haven't got someone you can share that with … (Spoken softly) Yeah it does make you sad. Which of course … detracts from your quality of life" (P16 – Partner).* The lack of interpersonal synchrony extended beyond simple companionship to the ability to express emotional understanding and support for one's partner:

I guess my mother is probably an example. I've been quite upset about her … her health is deteriorating … when I try to talk to him about it, ah, I don't really get any sense of him being particularly worried or caring (laughs) … I feel like maybe I would like some emotional support or something and (pause) he doesn't give any indication from his face. I suppose he does more from his actions, but in conversation he won't necessarily look as though he's particularly caring or concerned for me sorta thing. (P18 – Partner)

Although emotion may be absent from the faces of individuals with masking, there was no evidence in the interview data to suggest PDparticipants difficulty in displaying and reciprocating emotion reflected an underlying deficit in social or emotional understanding.

'It can affect you quite badly without you realizing exactly what it is': poor recognition and understanding of masking

Participants highlighted that an impairment to expressive behavior was important but difficult to recognize as a symptom of PD because it was inconspicuous: *"It's quite … probably one of the most serious things about the Parkinson's … when somebody tremors … you know this is Parkinson's … But with masking it's a bit subtle, and when you live with someone all of the time, it happens sort of, gradually, and you're not necessarily aware of it. So it can affect you quite badly without you realizing exactly what it is" (P14 – Partner).* Changes participants experienced in their relationships could also be difficult to associate with reduced social expressivity, which could lead to prolonged deterioration of relationship quality when symptom awareness is not forthcoming.

A further challenge was that many PDparticipants were initially unaware their facial expressivity or appearance had changed. Often family, friends and colleagues were first to note a reduction in the frequency or intensity of the PDparticipants smiling. Even when masking became clinically overt and was detected by observers, it was frequently misattributed to negative affect, or to other conditions such as stroke. Many partners and PDparticipants continued to be unaware facial expressivity could become impaired in PD after initial diagnosis of the condition, indicating a missed opportunity for identification of this symptom and the development of insight. The recognition of masking as a symptom of PD seemed to enable better adjustment to an expressive impairment, at least for some couples: *"I didn't have a clue … I blamed him a lot for it without (pause) understanding. So I felt that bad about that, you know. And the kids did too, but once they understood as well, it was like all that stuff went! The blame, the niggliness. All of it. It's interesting how you know, you think a diagnosis is disempowering, but often it's not. In our case it certainly hasn't been, it's been helpful" (P10 – Partner).* For others, gaining symptom knowledge provided explanation of their experience, but did not reduce their interpersonal and communicative difficulties.

'They don't get that information because it's not there and because the harm that's already done in a relationship isn't understood': unmet health resource needs

A principal factor in the low recognition and awareness of impaired nonverbal expressivity in PD may be the poor availability and quality of educative health resources. Only one couple reported receiving satisfactory education/support from a health practitioner regarding masking. Some participants sought out or came across online/written information, but generally found these resources did not go beyond simplistic symptom descriptions. All but one participant felt there was an unmet need for resources which focused on masking's wider implications: *"I mean, God, if people find it as tough as I do, or as tough as we do, as [my wife] and I do … people … definitely need something. Anything that can reduce, can help, even your partner, your friends, anything else, understand that you know, there is a reason why you look the way you do" (P03 – PDparticipant*). Self-management guidelines which had greater relevance to and emphasis on consequences for everyday social and psychological wellbeing were also desired.

One partner commented on the importance of education to make sense of the changes observed in the relationship and

prevent the formation of dysfunctional and inaccurate impressions of affect/behavior: *"the importance of information I think is, can't be underestimated… diagnosis, it gives you a context in which to make meaning of something. Whereas in the past you're kinda floundering around in a vacuum and making all sorts of misguided assumptions about what something is, based on your understanding of how someone's been, you know?" (P10 – Partner)*. The significance of information was further built upon by the following PDparticipant, who commented that the present lack of masking education and awareness could mean missing opportunities for a "recovery" of quality of life:

> *When people stabilize their medication you know, there's a sort of recovery there. But there's also a recovery… which no one's ever mentioned to me, where it comes with the understanding. Which is cool that it's there, but it's not so cool that people just don't get it… They don't get that information because it's not there and because the harm that's already done in a relationship isn't understood at all. (P02 – PDparticipant)*

Several participants expressed a desire for discussion of this complex and important topic which matched their needs, rather than a simple dispensing of health information. Specialist consultations and community support groups were suggested as appropriate contexts for such conversations. One participant recommended written and verbal information be provided by clinicians around the time of diagnosis, as part of a more holistic dialog about the influence of PD on emotional and social well-being, while accentuating opportunities for intervention across these domains. He had earlier emphasized a desire for health professionals to better informed of the implications of masking and to elicit if their patients are experiencing these difficulties:

> *Where the difference could have been really helpful was… for the GP to have understood more about it… to say, 'hey look, these are the things that could have been happening for you in your life and can potentially be a lot better'… I didn't understand the different aspects and… impacts at all …no one's talked to me about the masking thing, at all… if someone had been able to say, look there's a dopamine imbalance issue and you get a real emotional imbalance from that… there's emotional stuff and you might be feeling like this, and we can fix this. There's the facial stuff and that may have been f***king up your relationships for some time… without you realizing it, and people may be not reading you as you think they are. Have a think about that, but here's something to read about that… have sheet of paper… There should be a hand-out that people get you know. (P02 – PDparticipant)*

"Almost an asset": positive reappraisals of masking

Participants reflected upon the incidental benefits or positive change occasioned by adjustment to this symptom. Most participants were keen to point out masking was "almost an asset", because any benefit was qualified by the difficulties it presented. For example, appearing unreadable was used strategically to enhance workplace negotiations by two PDparticipants but in the context of close personal relationships, as one individual pointed out, *"it's quite hard to relate to people" (P03)*. Partners similarly stressed the mixed consequences of masking. While on the one hand masking created interpersonal difficulties, on the other it could be a catalyst for personal growth, fostering new communicative approaches and patience. For example, *"the whole thing has probably made me slow down a bit more, which has probably been a good thing you know because I've stopped to listen you know I've sort of asked [him] something I have to really listen to what he's telling is the answer. Because… it's gonna have to be a verbal description because the face won't tell me… But I s'pose that's probably been a good thing really you know" (P13 – Partner)*.

Supplementary analysis: compensatory behaviors and coping strategies

These responses were coded and organized into categories of compensatory and coping behaviors (summarized in Table 3). Heterogeneity was observed in individual coping knowledge and repertoire. A small number of individuals appeared to have adjusted well to living with masking and reported effective use of compensatory behaviors. Others described feeling helpless and reported minimal coping or compensatory tactics, while a few commented their present self-management approach was not overly helpful. Those that appeared to have adjusted well to an expressive impairment tended to report a lower severity of masking. However, the size of the cohort restricts further interpretation.

The findings highlighted the shared and diverging coping needs of people with PD and their partners. In general, partners reported their most used and useful strategies were those that challenged negative thoughts and feelings arising from misperceptions of masking, and enhanced communication or clarified misunderstandings. PDparticipants generally favored the use of speech, touch, and gesture to substitute for reduced nonverbal communication. Both partners and PDparticipants engaged in activities which fostered togetherness and emotional connection as a couple. Interestingly, just one participant regularly posed socially appropriate (voluntary) facial expressions to offset the loss of spontaneous expressivity. This participant was also the only female PDparticipant. Six other participants posed expressions infrequently, but this behavior was experienced as effortful and false, which prevented its regular use as a compensatory behavior. There may differences in the way women and men with masking experience and compensate for the loss of facial expressivity (but further interpretations cannot be made from the present study).

Discussion

This is the first qualitative study of facial masking in PD. The results extend previous findings demonstrating how stranger and health professional groups form unfavorable impressions of people who have masking [16–19]. The present study brought to light that partners of people with PD also tend to form negative perceptions of their loved one, misinterpreting people with PD's thoughts, feelings or communicative meaning. Compared with other aspects of the management of PD, this symptom may seem somewhat benign and of low clinical importance. However, this study highlights the significance masking has in the everyday lives of people who have PD and their families. Impaired expressivity led to social isolation, reduced emotional connectedness, relational detachment, and dysfunctional thought/behavioral processes (i.e., self-blaming causal attributions and social withdrawal). Participants described varying levels of distress associated with impaired nonverbal expressivity, which was exacerbated by poor awareness of this symptom and a perceived unmet need for patient education and support. At worst, the loss of facial expressivity had a profound effect on relationship quality and sense of wellbeing. Consequently, health and rehabilitation practitioners working with PD populations need to consider that this symptom may give rise to interpersonal and psychological difficulties. Practitioners should also recognize and respond to patient/familial needs for support and education.

Impaired facial expressivity appears to interfere with two processes crucial to human social cognition and interaction – the ability of partners to decode the emotions of people who have PD, and people with PD's ability to mirror or share in emotion with others. A key finding was that partners confused facial masking

Table 3. Summary of the compensatory and coping strategies for impaired nonverbal expressivity, used by the interview cohort.

Compensatory behaviors and coping strategies	Description/function	Participant examples (interview quotes)	Group	Number of participants
Questioning and checking in	Asking questions to compensate for the reduction of nonverbal behavior to confirm communicative intent, affect or psychological state.	*"I ask ... more questions to get, not the answer, but an answer ... .Whereas with other people- ... one question and a look will give you what you want to know."* *"Are you enjoying it? Are you happy?"*	Partners	7
Managing misinterpretations of affect	Tactics to reduce affect misidentification and address maladaptive cognitions. E.g. (1) inhibiting automatic evaluations through purposive orienting of attention towards non-visual communicate signals. E.g. (2) directly challenging the misidentification of negative affect and associated self-blaming/dysfunctional causal attributions.	*"You're kind of always watching really, if that makes sense. Always listening, because ... you can't see it so you have to hear what's being said or see what's being done or something to try and interpret."* *"I have to remind myself all the time that what I think he's feeling is probably not what he's feeling ... he's not unhappy because of me and kids ... And then just remind myself of all the good things that we are doing ... There's no reason for him to be unhappy sort of thing."*	Partners	5
Touch/Physical Affection	Used to compensate for reduced expressivity, maintain intimacy and/or minimize feelings of distance.	*"Probably touch and feel has probably taken the place of how I look or whatever, I think. Certainly, touching or cuddling her would, ah would be a way of me getting past the masking."*	PD participants	6
Verbalizing	Speaking aloud thoughts, intentions and emotions to augment the loss of facial behaviors and emotional expressions.	*"Quite often at work people look to me for approval and they're not getting it, even if I'm feeling it. So I verbalise it rather than rely on them reading my face"*	PD participants	4
Voluntary posing of socially appropriate facial expressions & bodily gestures	The conscious, effortful production of expressions and gestures congruent to felt affect or social interaction. For example, posing a smile.	*"As I've still got the ability to try and influence it, I would do that, rather than just let it sort of, compound as a grumpy face."* *"I express it so that it becomes, not just my face ... I'm saying it, I'm living it ... It's also contextualised by what's happening with the rest of my body ... so what's happening on my face doesn't look out of place ... It's the bigger picture"*	PD participants	Frequently (1), Infrequently (6)
	Self-administering additional anti-Parkinsonian medication to reduce PD symptoms *and* appear more socially expressive.	*"I usually end up taking at least maybe two extra tablets during the evening, just to put off the side effects and everything else, like ... both the memory and the masking as well"*	PD participants	2
Facial exercises	Physical exercises to preserve and improve the mobility of facial musculature.	*"I've got a sheet of paper that's got sort of exercises ... quite basic things, but I think they do make a difference, in keeping your muscles sort of supple."*	PD participants	Frequently (1), Infrequently (2)
Fostering closeness and togetherness as a couple	Interactions or activities which cultivate a sense of closeness and togetherness, outside of caregiving and other roles (i.e. parenting).	*"I think since the masking and that had more impact, we have had to try and make more occasions where it's just the two of us. Because ... we both need to put a lot more effort into our interactions ... we can't just take it granted that sitting side by side on the couch is going to give us that interaction now."*	Both	6
Adjusting to reductions in communicative/ expressive responsivity	Adjustment to masking and other impairments was described as requiring greater patience and compassionate thoughts and behaviors towards one's partner and for oneself.	*"I've learnt to, I have to give him time ... To (pause) um think or um articulate what he's thinking. It's taught me patience, which has never been a strong point of mine!"* *"We're really caring of each other and each other's vulnerabilities ... [she] cuts me a lot more slack now! (Laughter) ... She would never say, 'just go and have a lie down' before. She said, 'just get on with it you prick!'(Laughter) ... It's hard to separate stuff out, but a lot it is actually me lightening up on myself.*	Both	6
Educating others about facial masking and/ or PD	Explaining one's condition and educating others about PD. Sometimes by way of humor.	*"You still have to discuss and remind them that you know, that dad isn't grumpy with you and he may not look like he's having a good time, but he is having a good time."*	Both	5

with negative affect. Partners also made attributions about the cause of perceived affect, the nature of which could be distressing and dysfunctional. This may be because expressions are usually processed automatically and can be used to form judgments without intention or conscious awareness of having done so [28]. This suggests that partners and family members may benefit from assistance in identifying and challenging negative or distressing cognitions that arise when interacting with an individual who has reduced expressivity. Cognitive restructuring techniques may be particularly useful, as the thought processes partners described appear similar to common cognitive distortions such as mind reading and personalization [29]. For example, one participant described challenging automatic thoughts about her husband's unhappiness, by using a technique somewhat similar to

Table 4. Recommendations for the management of impaired nonverbal expressivity in Parkinson's disease.

Evidence/key findings	Suggested actions for psychosocial rehabilitation/ intervention	Example
Facial masking and its everyday consequences were not readily associated with Parkinson's.	Elicit insight of changes to facial appearance. Advise people with Parkinson's and families if this symptom is observed. Most crucially, discuss implications for everyday life.	Masking is frequently misread as negative affect or indifference. Relationships may become distant and unsatisfying – often without realization there may be an association to Parkinson's.
Masking caused substantial interpersonal and emotional difficulties for some individuals.	Monitor common markers of psychosocial distress/dysfunction in people with Parkinson's and their families.	Observe markers such as: frequent or distressing misinterpretations of negative affect, increasing relational detachment or disconnection, communicative difficulties and misunderstandings, avoidance of interaction.
Health provider support perceived to be insufficient. Patient resources reported to be of poor availability/quality. People with Parkinson's and their partners reported shared and differential coping tactics.	Educate and assist adoption of helpful coping and compensatory behaviors, matched to individual needs.	Assist partners to challenge assumptions arising from the misidentification of negative affect. Encourage people with Parkinson's to use speech, gesture or touch where possible, especially to show enjoyment, warmth and appreciation to loved ones.

Examining the Evidence [30], *"what I think he's feeling is probably not what he's feeling… he's not unhappy because of me and kids… remind myself of all the good things that we are doing… There's no reason for him to be unhappy sort of thing" (P12 – Partner).*

Participants described masking as a barrier to satisfying close relationships, as it could make individuals who had PD look uninterested, uncaring, and restrict emotional reciprocity. It seemed this was because a lack of facial expression was misread by others as negative affect, but could also be because the PDparticipants thoughts, feelings, and behaviors became more difficult for others to identify at all. This is consistent with previous research which proposes that people who experience masking may seem to be impassive, bored, unhappy or lack warmth [4, 14,15] and that observers may have difficulty seeing the "real person behind the mask" [4]. People automatically and non-consciously mimic one another's facial expressions; this process facilitates rapport and is important to empathy and interpersonal bonding [31]. Previous research has also demonstrated that people who have PD have decreased overall mimicry, with particularly weakened and delayed smiles [32]. Our findings suggest that the impairment of emotional expressivity might interfere with people with PD's ability to mimic others' emotion, and, therefore, feel or display empathy and emotional support. It may also be difficult for partners to feel close to their loved one who has masking because there is no observable emotion for them to readily mimic, or because they mimic perceived negative affect, in turn priming dysfunctional social behaviors such as avoidance and withdrawal. Partners also repeatedly emphasized the absence or reduction of positive affect, which is consistent with literature outlining the crucial role of nonverbal displays of positive affect in the maintenance of intimacy [9,10].

The lack of symptom recognition reported by interview participants is noteworthy. Many remained unaware of the presence of an expressive impairment for a prolonged time after PD diagnosis. It may be that this symptom is not recognized by people with PD and their families because of its low saliency relative to other signs of PD. In addition, prior research has indicated some people who have PD possess a limited awareness of the extent of their expressive impairment [33]. It is also likely this finding reflects a lack of knowledge that expressivity could become impaired in PD in the first place. The present study highlighted that current educational resources and health provider support for impaired nonverbal expressivity in PD might also be inadequate in New Zealand. This may indicate a gap in health practitioner knowledge of facial masking, or a focus on other aspects of the clinical management of PD. Research has previously demonstrated that General Practitioners (but not Specialists) in New Zealand may require training in providing PD information and becoming more responsive to this population's needs [34]. It is also possible people who have PD and their partners do not readily enquire about facial masking with their providers because of poor symptom insight or a reluctance to discuss interpersonal difficulties. These subjects are topics for future research.

Gaining an understanding of impaired expressivity appeared to somewhat mitigate the difficulties participants experienced. This would seem to suggest that early recognition and management of this symptom may enhance adjustment to an expressive impairment, preventing unnecessary deterioration of wellbeing. Participants in the present study desired more than a simple dispensing of information, indicating a need for informed health professionals and organizations who can facilitate dialogs on this topic as it becomes appropriate to people with PD and their families' needs. Given these findings, we recommend that health practitioners routinely elicit insight of people with masking and their families whenever this symptom is observed and provide practical educational/supportive interventions, where appropriate. Further implications for rehabilitation are presented in Table 4.

To the best of our knowledge, this study is the first to provide a description of how this unique population attempts to manage the loss of nonverbal expressivity, and to consider the role of psychosocial intervention in the management of masking. People with PD and their partners had shared, as well as differential, approaches to managing the interpersonal and psychological consequences of masking. Therefore, health professionals assisting PD populations to adjust to an expressive impairment should be aware of differing concerns and needs (see Table 4 for specific recommendations). Future scientific research is required to establish which strategies are the most effective and for whom. In other conditions (such as Moebius Syndrome), additional channels of communication may be available to offset the loss of expressivity. For example, using vocal tone to show emotion [13]. Little is known about which compensatory strategies are available and most helpful to individuals with PD. There is some evidence physiotherapeutic intervention may improve or sustain facial mobility/expressivity in PD populations from short-term rehabilitative programs [35,36]. Masking may also be reduced by speech therapy programs such as Lee Silverman Voice Treatment (LSVTR LOUD) [37]. Most participants in the present study were disinclined to voluntarily pose expressions to compensate for reduced

spontaneous expressivity. Posing expressions was perceived to be overly effortful and inauthentic, or participants believed they had insufficient voluntary facial movement to approximate expressions. This somewhat aligns with previous findings where posed smiles of people who had PD were found to appear phoney [38]. We suggest that health practitioners encourage a facial mobility plan but be aware people with PD may prefer rehabilitation approaches which facilitate positive social function over the maintenance of facial motor movement. Until evidence demonstrates the effectiveness of different compensatory strategies for facial masking, health practitioners may wish to utilize those outlined here as a general guideline, to educate people who have PD and their families in adopting helpful coping tactics (see Table 3).

It has recently been proposed that social factors influence the psychological difficulties people with PD face and actively disable or limit their wellbeing [39]. Our findings are consistent with this perspective. For example, PDparticipants were mocked for their unmoving facial appearance, and social participation could be restricted when PDparticipants experienced negative thoughts about other's impressions of their reduced expressivity. One way to address the social barriers people with PD experience could be to raise awareness of this highly visible but misunderstood symptom, within general and PD populations.

This study had several limitations. It would have been interesting to include more female participants with PD, as some researchers speculate the loss of expressive capacity would violate social norms more for women than men [40]. The ethnic homogeneity of the cohort suggests the interview participants may not be typical of all people who have PD in New Zealand. This study included nine couples who self-selected and may over-emphasise the level of distress this symptom produces for PD populations. All interview data were analyzed together, without focus on participant status. Future research using patient and spousal dyads may bring to light different or novel findings. Data from the face-to-face interviews may be emphasized in the analysis, because the telephone interview format allowed less opportunity for the interviewer to develop a sense of familiarity with the interviewee. Interviewer observations of masking were not analyzed and could have offered additional data. Participants self-reported masking, which was verbally confirmed during eligibility screening. It was not practicable to formally assess participants for the presence and magnitude of expressive impairment. This may have provided additional data of interest. Future research is required to confirm the novel findings and recommendations presented in this study.

Conclusions

PD is a progressive and incurable condition. It has diverse functional, cognitive, psychiatric, and social consequences. Within chronic conditions like PD, the quality of close relationships is significant because of the well-established association between social support and both physical and mental health [41]. Preserving social-emotional functioning may be especially crucial in PD, where care is often provided in the home and participation in other meaningful activities becomes increasingly restricted, to sustain quality of life. Because of the expected rise in age-related conditions like PD, disease management knowledge is poised to be of increasing importance. Health and rehabilitation practitioners working with Parkinson's populations should consider facial masking as an etiology of interpersonal and psychological difficulties and be responsive to individual and family needs for greater support and education.

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5. Study 2
Development and Validation of a Novel Instrument for the Assessment of Hypomimia Impact in Parkinson's Disease

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Development and Validation of a Novel Instrument for the Assessment of Hypomimia Impact in Parkinson's Disease.

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ABSTRACT

Objective: Facial masking is a characteristic symptom of Parkinson's disease, traditionally measured by determining symptom severity. Existing measures are based on clinicians' observations and may not address experiences of living with facial masking day to day. We report the development and validation of two self-report questionnaires for use with people with facial masking and their significant others.

Method: Questionnaire items were generated from in-depth interviews with people self-reporting facial masking, their partners, and relevant literature. Expert opinion and feedback from members of the interview group were used to refine the provisional items. Preliminary questionnaires were administered to a separate sample (N = 138) for psychometric evaluation.

Results: The Parkinson's version of the questionnaire (n items = 28) measures facial masking severity and impact, comprising individual and relationship difficulties subscales (n = 6 each). The Significant Other version is a unidimensional measure (n items = 30), with a single facial masking difficulty subscale (n = 18). Internal and test-retest reliability were good (Parkinson's version subscales α = .830, r =.762, and α =.765, r = .729, Significant Other α =.936, r = .895). Convergent validity was supported by satisfactory correlations with scales measuring emotional health, relationship satisfaction, quality of life, and caregiver strain (Parkinson's version all r's > .25, Significant Other all r's > .26, $p < 0.05$).

Conclusion: The Facial Masking Questionnaire provides assessment of socioemotional consequences of FM. Psychometric evaluation showed the questionnaires are reliable and well-accepted by people with facial masking and their significant others.

Parkinson's disease (PD) is a chronic and progressive neurodegenerative condition characterized by difficulty with motor-movement.[1] It has a negative impact on subjective wellbeing, and as PD is common in middle to older age, poses an increasing burden on aging societies[2]. Since research into PD treatment is ongoing, increasing attention has turned to improving the lives of people with PD and their families. This article describes the development of a tool to assess and monitor the social and psychological consequences of facial masking in PD.

Facial masking (FM), variously known as Hypomimia, facial bradykinesia, or the mask of Parkinson's, is the reduction of facial emotional expressivity. It arises from motor deficits which occur in the muscles of the body in PD, including the muscles of the face. FM can result in an unusual stillness of the face and reduces the capacity to express emotions and convey thoughts and intentions[3]. FM may be in present in 70% [4] to 90% [5] of people with PD, but its impact on the experience of everyday life in PD is poorly understood.

The bulk of existing research focuses on the influence of FM on the social impressions of strangers[6-8] and healthcare practitioners.[9-11] These studies find that even experienced healthcare practitioners are not immune to the influence of FM and form a negative impression of those with FM. However, research has largely overlooked the impact of living with FM for people with PD and their families. The consequences of having FM, or having a loved one with FM, remain largely unexplored.

Two assessment tools offer evaluation of FM status in PD (MDS-UPDRS and ICRP-PD)[12, 13] yet no self-report questionnaire to measure FM-related psychosocial difficulties has been developed. Quality of life instruments for PD[14-18] also omit specific reference to FM. Existing measures typically assess FM severity focusing on blinking, active expressivity in the face, and active lip/mouth closure during speech.[12, 13] These methods are useful for research or clinical staging, rather than evaluating the impact of having FM day to day.

There are several reasons that justify the development of an assessment tool measuring FM impact. Foremost, evidence shows the negative consequences of FM extend beyond casual or healthcare interactions to close relationships. A greater sense of stigma

associated with having FM, and being female, are found to predict poorer quality of life in people with PD.[19] FM also adversely impacts relationship enjoyment for PD care partners.[20] Secondly, because PD treatment aims to lessen the impact of the condition on people's lives[21, 22], addressing problems associated with FM is a worthwhile undertaking. The development of a measure will help to generate insights about the concerns and needs of individuals living with FM. Indeed, some individuals have poor FM awareness or may not bring up this topic with their healthcare providers, and many reported a desire for increased access to FM education and support.[23] This is not surprising given the challenges of providing information in a complex condition like PD.[24, 25] However, it is important that clinicians who work with PD increase their own and their patients' awareness of the psychosocial consequences of FM. Thus, our intent in developing a new measure was to aid understanding of FM's consequences in close relationships, and to provide a structured way for healthcare professionals to identify individuals that need help or support with this symptom.

This paper describes the process of questionnaire development and initial validation among a community sample of people who have Parkinson's and their significant other (partner, close friend, or family member aged over 18 years). The first objective was to generate items for the preliminary questionnaire and evaluate its content validity, general feasibility, and clinical applicability. Secondly, we examined the reliability and construct validity of the questionnaire.

MATERIALS AND METHODS

Item Generation & Scale Construction

This questionnaire aims to capture perceptions of the extent of expressive impairment and rate the amount of emotional and social difficulty associated with FM. As FM effects individuals who have PD and the people around them, separate versions of the questionnaire were developed for people with PD (PwP) and their significant others (SO, partner/spouse, adult family member, or close friend). Although item content varies, each version covers similar dimensions associated with FM: psychological characteristics or distress, and difficulties in close relationships (including relationship quality).

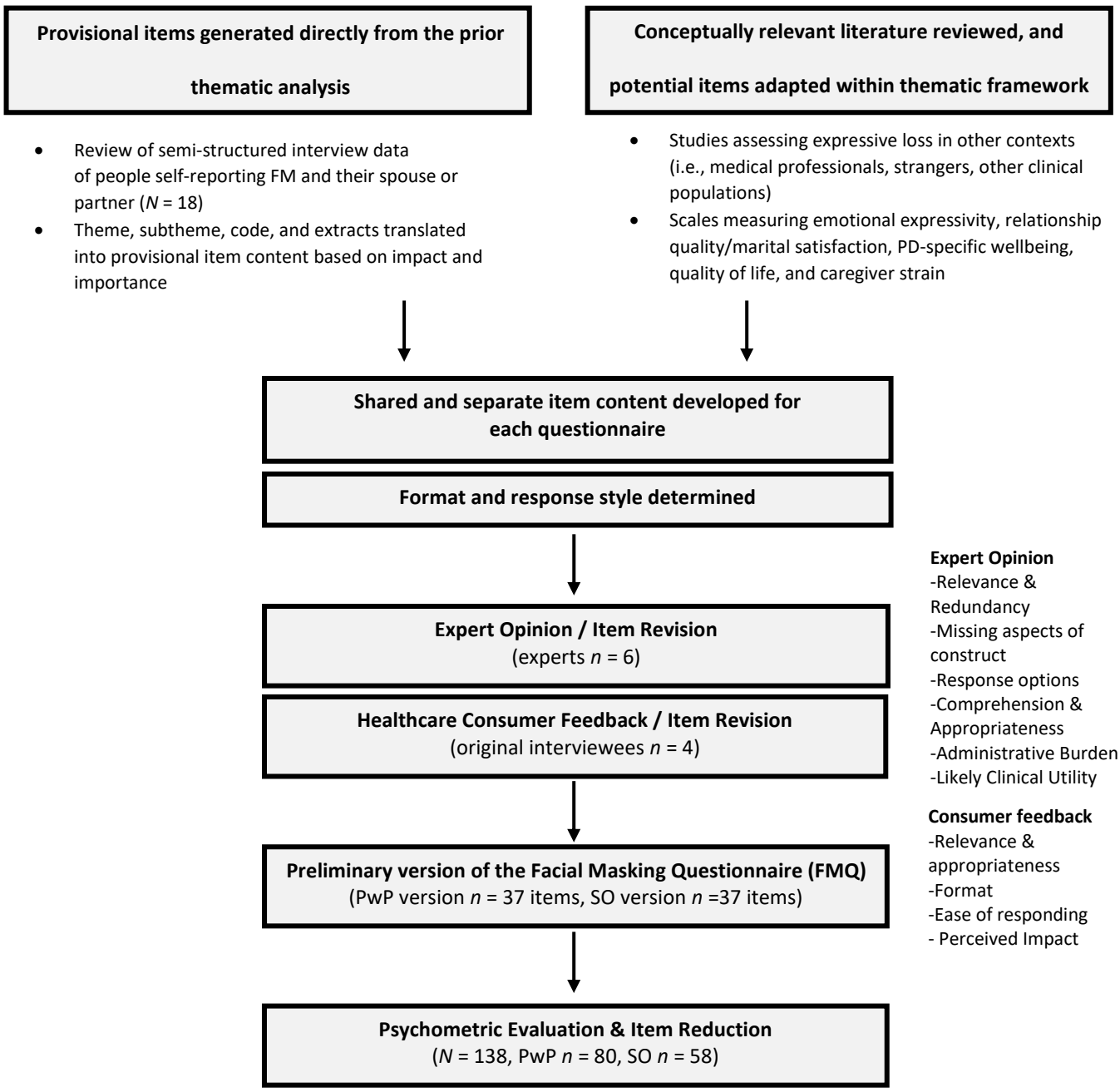
Figure 1 summarizes the development of The Facial Masking Questionnaire (FMQ). Scale construction followed the steps outlined by DeVellis[26] and Creswell & Plano Clark.[27] To incorporate experiences of FM in everyday life, items were primarily generated from semi structured interviews with people with PD who self-reported FM and their partners.[23] Existing literature was reviewed for conceptually relevant variables that could be adapted to fit the experiences of individuals with FM and their partners. Expert opinion and healthcare consumer feedback was used to refine the preliminary items. This was followed by psychometric evaluation and item reduction of the questionnaires. A more detailed description of the development process and preliminary items is provided in the Supplemental Data Appendix I.

The preliminary FMQ-PwP and FMQ-SO each contained 37 items (Supplemental Data Appendix I). The first two items asked individuals to rate the severity of FM on a 4-point scale (#1), and to rate knowledge their knowledge of FM (#2). This was followed by 28 items assessing emotional and social difficulties associated with FM in close relationships. Items (#3-30) were rated by selecting how often each statement occurred/applied to them on a four-point, Likert style response scale (*Never or Rarely, Sometimes, Often, Almost all the time*).

Participants were then asked three questions to screen for psychological health *(I feel uncertain, worried, or concerned; I feel angry, irritated, or frustrated; In general, I feel happy or in good spirits*) using the same Likert response scale. This was followed by two open ended questions asking about strategies they use for coping with FM, and to describe their experiences of living with FM. The last two questions asked participants to rate the overall FM-related difficulty on a 5-point scale (#36), followed by a yes/no item asking if they would like further information and support (#37).

Figure 1

Flowchart summarizing the development and validation of The Parkinson's Facial Masking Questionnaire (FMQ-PwP and FMQ-SO).



Validation of the Facial Masking Questionnaire

Design and Setting

The questionnaires were validated in a cross-sectional multiple center study that included 138 participants. This comprised people with PD (PwP, *n* = 80) and their significant other (SO, *n* = 58) who came from a general community sample across different regions of the North Island of New Zealand (NZ). Several waves of advertisements seeking participants were placed in PD support group newsletters and emailed directly to support group members. Data collection took place throughout March to November 2019.

After potential participants made contact, they were telephoned and screened for eligibility. Potential participants were provided with verbal/written information about the study, before being asked if they consented to taking part. Testing took place at participant's home or workplace. At the conclusion of testing, participants were asked if they were happy to take part in retesting in approximately one month's time.

Inclusion and Exclusion Criteria

Individuals with a diagnosis of idiopathic PD were eligible to participate. Those who self-reported diagnosed cognitive impairment or who had difficulty conversing in English, were excluded. Individuals with PD but who did not have a significant other were eligible to participate. Selection was *not* made on the basis of FM status, so that participants had a range of FM severity from none to severe.

Assessment

Demographic and disease-related variables were collected first. The two Facial Masking Questionnaires (FMQ-PwP & FMQ-SO) were administered alongside other self-report scales assessing health and wellbeing. Individual Expressive Disposition was assessed using The Berkeley Expressivity Questionnaire (BEQ).[28] A modified BEQ was also created for SO participants to complete. Affection and enjoyment dimensions of social relationships were measured using the affectionate support and positive social interaction

subscales of the Rand Medical Outcomes Study – Social Support Survey (MOSS-SSS)[29], which has previously been used in people with PD and their partners.[20] Participants whose SO was a marital or long-term partner rated relationship satisfaction using the Kansas Marital Satisfaction Scale (KMSS).[30] To measure psychological health, PwP completed the Hospital Anxiety and Depression Scale (HADS)[31], which has been found valid in PD populations.[32] Quality of life for PwP was assessed with the Health-related Quality of Life Instrument: Fifteen dimensions (15D).[33] SO quality of life was assessed using the World Health Organization Quality of Life Assessment -Short Version (WHOQOL BREF)[34]. Strain related to caregiving role in SO participants was measured using the Caregiver Strain Index (CSI).[35]

Statistical analyses

All analyses were calculated using IBM SPSS (version 26 for windows). Demographic and disease-related variables are presented descriptively. PwP and SO versions of the Facial Masking Questionnaire were analyzed separately. Psychometric analyses focused on the main scale items (#3-30). Items with occasional (< 15%) missing values were replaced with item means for analysis, because losing further data in this relatively small sample could influence the generalizability of the analysis and bias factor loadings.[36]

Items were assessed for suitability in a number of different ways including, no more than 15% of missing data, the use of a range of response options (≥ 3),[37] item mean within ±1 of the mid score, item mean within ± 0.50 of the median, and item standard Deviation > 0.75. Floor and ceiling affects were examined, with items scrutinized case by case basis to consider items that did not meet the above requirements but focused on conceptually or clinically important information for some individuals.[38] Such items were excluded from the factor analyses as they are likely to produce uninterpretable factor[390] but were retained in the final questionnaires (see Table 2).

The dimensionality of the main items was examined using Exploratory Factor Analysis (EFA). Since the objective was to explore the underlying structure of a novel rating

scale where little is known a priori, screening addressed the overall factorability of the data, rather than data reduction. This involved checking conceptually related items were clustered together in the range of 0.2 to 0.9.[38] Principal Axis Factoring was performed, and scree plot inspection and Kaiser's Criterion (eigenvalues ≥1) were used to identify interpretable factors. Sampling adequacy (Keiser-Meyer-Olkin statistic), sphericity (Bartlett's test), and anti-image matrix diagonal/off diagonal values were checked.

The internal consistency of items comprising each factor was tested by Cronbach's alpha coefficient. An alpha of > .7 [40, 41], mean inter-item correlation range of .15 to .50 [42], and item-subscale correlations of > .4 [43] were taken as acceptable.

Test-retest reliability of the questionnaire subscales was evaluated using an Intraclass Correlation Coefficient (ICC, two-way random effect model with absolute agreement)[44], and evaluated according to guidelines for psychological instruments.[45]

Convergent validity was examined by comparing correlation coefficients between the scores on the FMQ with scores on rating scales measuring related wellbeing constructs. Criterion validation could not be conducted for the main items, as no corresponding measure could be located for comparison. The extent to which FM-severity rating (a single index item) corresponded with the Berkeley Expressivity Questionnaire, an existing measure rating individual expressive disposition, was examined.

Approval and consent

Ethical approval for this study was received from the University of Waikato Human Research Ethics Committee, prior to commencement (ref#2018:46). Respondents received verbal/written information about the study and gave their informed consent before participating. Participants received a $20 supermarket voucher to thank them for taking part.

RESULTS

Demographic and Disease-Related Data

A cohort of 138 participants (PwP *n* = 80, SO *n* = 58) were included in the sample. The demographic and clinical characteristics for the participants are displayed in Table 1.

Psychosocial Consequences of FM

The right-hand column of Table 1 shows the mean FMQ scores by self or SO-reported level of FM; 66.25 % (*n* = 53) of PwP reported themselves to have FM, and 86.21% of SO (*n* = 50) reported FM. Most participants categorized FM severity as low or moderate (PwP 63% and SO 81%). For both PWP and the SO, those reporting higher levels of FM severity obtained higher total scores on the FMQ. A similar pattern was observed for the other health and wellbeing scales; those with more severe FM had more anxiety/depression symptoms (HADS: PWP), lower marital satisfaction (KMSS: PWP & SO), increased caregiver strain (CSI: SO), lower social support (MOS-SSS: SO) and poorer psychological health (WHOQOL BREF: SO) [data not shown for brevity].

Table 1

Sociodemographic and clinical characteristics of study participants

PwP Participants (n=80)			
Demographics		**Health and wellbeing measures**	mean (SD) range
Age in years mean (SD) range	69.64 (9.36) 44.00-86.00	MOS-SSS Total Score	7.94 (1.58) 4-10
Male n (%)	57.00 (71.25%)	KMSS Total Score	19.36 (2.34) 12-21
Early Onset < 50 years n (%)	6.00 (7.50%)	HADS Total Score	10.16 (6.04) 0-29
Time since diagnosis mean (SD) range	6.40 (4.99) 0.50-22.00	15D Total Score	.734 (.09) .47- .93
FM self-reported at eligibility n (%)	60 (75.0%)	FMQ -PwP Total Score	
Dopamine Replacement n (%)	74.00 (92.50%)	Self-rated FM None	4.93 (3.25) 0-11
No medication n (%)	4.00 (5.00%)	Self-rated FM Low	7.88 (3.71) 2-17
Deep Brain Stimulation n (%)	3.00 (3.75%)	Self-rated FM Moderate	12.22 (7.48) 3-27
New Zealand or Other European n (%)	73.00 (91.25%)	Self-rated FM Marked	20.67 (5.51) 15-26
Māori & Pasifika n (%)	5.00 (6.25%)		
Other ethnic background n (%)	3.00 (3.75%)		
Retired or unable to work n (%)	65.00 (81.25%)		
Married/Living together n (%)	68.00 (85.00%)		
Took apart alone n (%)	22.00 (27.50%)		
Lives in private home n (%)	73.00 (91.25%)		
SO Participants (n=58)			
Demographics		**Health and wellbeing measures**	mean (SD) range
Female n (%)	45.00 (77.59%)	MOS-SSS Total Score	7.58 (2.02) 2-10
Relationship length mean (SD) range	41.97 (12.52) 11.00-62.00	KMSS Total Score	1.24 (4.13) 3-21
New Zealand/Other European n (%)	55.00 (94.83%)	WHOQOL Psychological Health	14.37 (1.96) 9.33 -17.33
Other ethnic background n (%)	2.00 (3.45%)	WHOQOL Social Relationships	15.37 (3.09) 6.67–20.0
Māori n (%)	1.00 (1.18%)	CSI Total Score	3.55 (3.17) 0-10
Retired n (%)	40.00 (68.97%)	FMQ -SO Total Score	
Has current illness n (%)	9.00 (15.52%)	Self-rated FM None	3.75 (3.37) 1-10
Tertiary Education n (%)	31.00 (53.45%)	Self-rated FM Low	12.03 (8.68) 0-38
Took part with spouse/partner n (%)	55.00 (94.83%)	Self-rated FM Moderate	16.79 (10.03) 1-33
Took part with parent (n, %)	3.00 (5.17%)	Self-rated FM Marked	39.33 (8.51) 33-49

Note. Dopamine Replacement, Sinemet or Madopar use. MOS-SSS, Rand Medical Outcomes Study – Social Support Survey; Affectionate Support and Positive Social Interaction Items only; KMSS, Kansas Marital Satisfaction Scale; HADS, Hospital Anxiety and Depression Scale; SO Psychological and Social Relationships Subscales, WHOQOL BREF, World Health Organization Quality of Life Assessment -short version; CSI, Caregiver Strain Index; 15D, Health-related Quality of Life Instrument -Fifteen dimensions; FMQ, Facial Masking Questionnaire, self-reported level of FM severity.

Psychometric Properties of the FMQ

Item Level Descriptive Statistics and Acceptability

Four items were excluded from the FMQ-PwP. Two items each were excluded for frequent missing values and low response variability among respondents with FM. For the FMQ-SO six items were removed, two items for frequent missing values, three items for low response variability among FM-reporting respondents, and one item for insufficient inter-item correlations. Details of all excluded items are provided in the Supplemental Data Appendices II and III.

Dimensionality

A principle axis factor (PAF) analysis was conducted with orthogonal (varimax) rotation for the FMQ-PwP, as initial analyses revealed factors were not correlated.[37] Nine items were eliminated from the analysis as they did not contribute to the factor structure, with content of removed items mostly reflecting relationship quality. One item each was removed for low loading (<.4) and cross loading (absolute difference in factor loadings < .2). Seven items were removed for low communalities (<.2).

A scree plot and Eigenvalues of > 1 identified two major factors and several minor factors. A two-factor solution was preferred to aid interpretability. The PAF was run a second time to check if the factor structure held following reliability analysis (where a further three items were removed for relatively high item total correlations > .7, or to improve reliability).

Table 2 shows the final factor matrix of the FMQ-PwP (*n* =12 items). Sampling adequacy (KMO = 0.717), Sphericity ($p < 0.001$), and anti-image diagonal/off-diagonal values met predetermined criterion. A two-factor solution was retained with eigenvalues of 3.2 and 2.6, which combined accounted for 44.03% of the variance.

For the FMQ-SO, PAF analysis was conducted (*n* items = 22). All items contributed satisfactorily to the factor structure. Scree plot inspection and Eigenvalues of >1 identified one major factor and three minor factors. Solutions for 1 to 4 factors were examined, with a

single factor solution the most clinically interpretable. The PAF was run a second time to check if the factor structure held following reliability analysis (where four items were removed for relatively high inter-item correlations >.7, or corrected item totals > .8).

Table 2 shows the final factor matrix for the FMQ-SO (*n* =18 items). Sampling adequacy (*KMO* = 0.842) and Sphericity ($p < 0.001$) met predetermined criterion. A single factor solution was retained with an eigenvalue of 7.50, which accounted for 41.67% of the variance. Although no items correlated >.80, the determinant of the matrix (0.00005) and residual off diagonal values suggested further redundancy in the scale (possibly due to the relatively small sample size).

Table 2

Factor Loading Matrix of FMQ main scale items and clinically or conceptually relevant items retained outside main scales future testing.

FMQ – PWP (n=12 items) *	Coefficients Factor 1	Coefficients Factor 2	Communalities	% Variance	α
8. It troubles me that way I feel inside is different from how I look on the outside	**.849**	-.086	.722	26.70	.830
7. It troubles me that my face does not look like me	**.778**	-.057	.623		
11. I withdraw from people close to me because of my facial appearance	**.727**	.126	.543		
14. I avoid interactions with others because of what they might think of my face	**.681**	.147	.484		
5. When I make an expression like a smile, it takes a lot of effort, feels false, or "put on"	**.660**	.099	.442		
4. People comment I appear to be in a bad mood much of the time	**.431**	.189	.216		
15. (R) My partner listens to me	.036	**.748**	.627	17.33	.765
9. (R) My partner is supportive and understanding when I need to talk or confide in them	-.070	**.721**	.419		
22. (R) I feel loved and wanted by my partner	-.026	**.576**	.348		
21. My lack of expression creates a barrier with my partner	.177	**.513**	.333		
18. My partner avoids or ignores me because my face does not respond	.165	**.505**	.319		
20. Interaction with my partner can feel uncomfortable or stilted	.109	**.473**	.208		

FMQ – SO (n=18 items) **	Coefficients	Communalities	% Variance	α
Q19. I feel disconnected from my partner because their face does not respond to me	**.794**	.630	41.67	.936
Q29. (R) My partner makes me feel loved and wanted	**.742**	.551		
Q3. My partner appears to be in bad mood a lot of the time	**.737**	.543		
Q10. (R) I can count on my partner to be supportive and understanding	**.737**	.543		
Q8. I find myself second-guessing what my partner is thinking and feeling	**.733**	.538		
Q4. It is hard for me when my partner's face does not respond	**.729**	.531		
Q27. I wish I were closer to my partner	**.683**	.467		
Q16. It is upsetting my partner's face/expression has changed so much from their former self	**.676**	.457		
Q11. I feel rejected or criticized when my partner shows little expression	**.671**	.450		
Q15. (R) I have a close and warm relationship with my partner	**.653**	.427		
Q24. I think my partner might be better off without me	**.624**	.389		
Q5. (R) My partner shares in my life's ups and downs	**.570**	.325		
Q22. (R) I feel a strong emotional connection with my partner	**.566**	320		
Q12. (R) My partner is interested in interacting with me	**.546**	.299		
Q6. (R) I can easily tell when my partner is feeling positive emotions	**.540**	.291		
Q9. I think my partner's unhappiness must be my fault	**.525**	.275		
Q17. I remind myself how my partner's face looks on the outside is probably not how they feel inside	**.495**	.245		
Q25. I worry my partner cares less for me than before	**.469**	.220		

Items retained outside main scales for clinical or conceptual importance

FMQ – PWP	FMQ – SO
3. When I am happy people close to me can easily tell what I am feeling.	7. My partner is more expressive with other people than with me
10. My partner misreads my emotions, attitude, or communication	14. I avoid being near my partner or interacting with my partner
12. People have been unkind to me because of my facial appearance	20. My partner's emotional signals make me concerned they might want to end our relationship
13. My partner can usually tell what I am feeling	21. My partner's emotional signals make me concerned they are considering an affair or are having an affair
16. I feel isolated from people important to me because they cannot see emotion on my face.	23. My partner's staring or lack of expression gets in the way of our sexual relationship
17. I take extra medication to make my face more expressive in social situations.	26. My romantic relationship with my partner is strong and rewarding
26. My romantic relationship with my partner is strong and rewarding.	

Note.: EFA results presented here verified following item reduction with reliability analysis. Major loadings in bold.

* PAF with varimax rotation, retaining a two-factor solution.

** PAF retaining a single factor solution.

Internal and Test-Retest Reliability

Cronbach's alpha was good for both questionnaires (Table 2). For the FMQ-PwP Individual Difficulty subscale (n = 6 items), internal consistency was acceptable (α = .830), however the mean inter-item correlation (r = .548) fell marginally outside the recommended range. For the FMQ-PwP Relationship Difficulty subscale, the internal consistency of the final items (n = 6) was acceptable (α =.765). The average inter-item correlation (r = .364) was within recommended range and item-scale correlations were acceptable (> .4).

On the FMQ-SO FM Difficulty Scale, the internal consistency of the final items (n = 18) was acceptable (α = .936). All item-scale correlations were high (> .4), and the average inter-item correlation was within recommended limits (r = .461).

A good to excellent degree of test-retest reliability was found between the subscale across two administrations, for both versions of the questionnaire (Table 3).

Validity

Correlation of the FMQ-PwP total and subscales scores with other rating scales measuring related constructs of psychosocial wellbeing was weak to moderate (Table 4). The FMQ-SO total and scale scores had weak to strong correlations with other rating scales measuring psychosocial wellbeing. Construct validity was supported by the overall pattern of correlation, which was as expected for questionnaire measuring a related yet distinct phenomenon from mood difficulties, social support, caregiver strain, quality of life, and relationship satisfaction (see Table 4).

Criterion validity of the FM severity item on the FMQ-PwP with total expressive disposition on the BEQ was weak (r = -.233), and not significantly correlated with individual BEQ subscales. In comparison, criterion validity of FM severity on the FMQ-SO with BEQ (modified SO version) was moderate (Expressive Disposition Total r = -.555, Negative Expressivity Subscale r = -.434, Positive Expressivity Subscale r = -.517 and Impulse Strength Subscale r = -.334).

Table 3

Test-retest reliability coefficients, means, and standard deviations for main subscales of the FMQ

	Test	Retest	Test-Retest Reliability (average measurement)			(single measurement)				
Measure Subscale	**M ± SD (range)**	**M ± SD (range)**	**ICC**	**Lower**	**Upper**	**ICC**	**Lower**	**Upper**	**F**	**P**
FMQ-PwP Individual Difficulty Subscale	3.37 ± 3.37 (0-15)	2.96 ± 2.98 (0-11)	.885	.784	.916	.762	.645	.844	7.445	.001
FMQ-PwP Relationship Difficulty Subscale	1.75 ± 2.44 (0-12)	2.07 ± 2.75 (0-13)	.843	.747	.903	.729	.597	.823	6.387	.001
FMQ-SO FM Difficulty Scale	11.1 ± 9.13 (0-40)	13.0 ± 10.29 (0-38)	.945	.890	.971	.895	.802	.943	20.681	.001

Note. Includes data of participants who completed the retest outside of 4-weeks due to personal or medical circumstances.
Test-retest interval (PwP, days) M=37.93, SD=13.76 & range= 18-108, (SO, days) M=39.00, SD=14.63 & range = 18-108.

Table 4

Correlation of FMQ subscales and total scores with other measures of psychosocial wellbeing

FMQ- PwP	HADS Total	HADS Depression	HADS Anxiety	KMSS Marital Satisfaction	MOS-SSS Total	MOS-SSS Affectionate Support	MOS-SSS Positive Interaction	15D HR QoL
Individual Difficulty Subscale Score	**.381****	**.429****	**.271***	-.023	.014	-.007	.087	**-.260***
Relationship Difficulty Subscale Score	**.303****	**.281***	**.251***	**-.455****	**-.394****	**-.431****	**-.256***	-0.190
Total score [a]	**.406****	**.454****	**.282***	**-.337***	**-.279***	**-.263***	-.188	**-.349****

FMQ- SO	CSI Caregiver Strain	WHOQOL BREF Psychological Health	KMSS Marital Satisfaction	MOS-SSS Total	MOS-SSS Affectionate Support	MOS-SSS Positive Interaction
FM Difficulty Scale Score	**.530****	**-.262***	**-.505****	**-.328***	**-.301***	**-.269***
Total score [a]	**.539****	**-.277***	**-.524****	**-.344****	**-.315***	**-.284***

Note. Spearman rank correlation coefficients. [a] Includes subscale scores plus clinically or conceptually important items excluded from the main analysis due to inadequate psychometric properties.
*Correlation is significant at the 0.05 level (2-tailed).
**Correlation is significant at the 0.01 level (2-tailed).

DISCUSSION

This paper provides an alternative approach to existing assessment of FM, which focuses on clinician-based outcomes. The FMQ measures the consequences of FM for psychosocial functioning, turning attention to the impact of *having* FM for people who have Parkinson's and their families. The results suggest the FMQ is a well-accepted and reliable tool with initial support for validity, which can serve to support management of FM in PD.

An additional advantage of the FMQ is the inclusion of peer-report (SO) version. The wellbeing of partners is of special importance in the clinical management of PD, since practical day-to-day care is commonly provided by family members in the home[46] and because of the close interconnection between caregiver strain and health outcomes for individuals with PD.[47]

Respondents who perceived greater FM severity tended to obtain higher FM Difficulty scores on the FMQ and poorer subjective wellbeing ratings on other measures. Whilst earlier studies showed the relationship between FM and quality of life to be mediated by stigma[19], our interview and testing samples seemed less concerned about feelings of stigma in the context of their closest relationships. However, responses to the FMQ open answer items suggested some participants did feel self-conscious of FM in public places or at social gatherings, such as at work, church and on the marae (cultural sacred place). Future investigation of the influence of FM and the factors mediating its impact should focus equally on social interactions in groups or with casual acquaintances, and on more private relationships with immediate family and friends.

Individual and relationship difficulties were not significantly related for PwP in the FMQ, and many general relationship quality items were removed during the questionnaire development. For SO, the most interpretable solution unified individual and relationship dimensions as single theme. This suggests that there is a difference in perspectives of the impact of FM between PwP and SO's and is consistent with previous findings where higher

FM severity was associated with decreased relationship enjoyment in partners, but not people with PD themselves.[20]

The low correlation of the FM severity item in FMQ-PwP with external measures of expressive disposition was not surprising. Whilst awareness of expressivity deficits has been shown in other PD populations[48], about 13% of our PwP participants (*n* =10) reported they had never heard of FM, and 19% (*n* = 15) were not sure if they had FM before testing. Low awareness of FM in PD and a failure to attribute related relationship problems to FM, were also salient themes of our original qualitative interviews.[23] This may mean some individuals who experience FM-related problems are not able to be identified by the FMQ, as the items assume respondents have a degree of symptom awareness. Despite this, supporting patient awareness and insight about FM is an important topic.

Psychometric properties of the FMQ were satisfactory, but a key limitation of this study is the sample size. This reflects, in part, the relatively small NZ population and the lack of a central register for PD. Further validation of the FMQ in a larger and separate sample is required.

As relatively little research on FM impact has been conducted, other aspects of experienced FM may not be captured by the FMQ. Further research in this area might explore fears about the meaning of losing emotional expressivity, perceptions of how loved ones versus casual acquaintances react to FM, and feelings of stigma in public places or social gatherings (such as work or when meeting friends in public). Nonetheless, as no current measures assess the psychosocial consequences of FM, the FMQ is a promising measure that will be useful for clinicians and as a prompt to raise awareness of FM.

CONCLUSION

The FMQ extends existing assessment of FM by focusing on the everyday consequences of FM for individuals with PD and their families. Whilst further validation of this tool is recommended, it will be of interest to clinicians who wish to take better account of the social and emotional challenges of FM, by identifying key concerns and needs for help. We hope this study will foster a greater appreciation of this unique aspect of PD.

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Data Availability

Due to the nature of this research, participants of this study did not agree for their full data to be shared publicly, so supporting data is not available due to ethical restrictions.

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Supplementary Data

Study 2 Appendix I. Development of the Facial Masking Questionnaire (FMQ)

Review of Qualitative Data

Provisional item content was generated based on interviews with nine couples who self-reported FM (interviewees *n* = 18), from our previous qualitative work on experiences of FM (see Wootton, Starkey, and Barber, 2018). At that time (early 2016), we could not locate any other study which investigated FM from the standpoint of people with PD. Therefore, the themes, subthemes, codes, and quotes from our qualitative findings were closely examined to develop item content. This was not a strict translation of thematic content into items but considered the impact and importance of the various thematic elements. We focused on examining the thematic data for the most salient and important problems described by people with FM and their partners. Where possible, provisional items were phrased similarly to the interview data. Distinct and overlapping themes across the person with PD (PwP) and Significant Other (SO) groups led to the development of shared and differential items for inclusion in the two questionnaires. A small number of items were included about social difficulties in less familiar contexts, such as having colleagues or strangers be unkind about a visible facial difference.

Review of Existing Literature

Existing assessment literature was reviewed for topics that were not covered by the interviews but could be conceptually relevant, with potential item content collated and adapted to fit the experiences of individuals with FM and their SO. We located conceptually related measures falling into two broad categories: the measurement of individual emotional expressivity and/or FM severity, and the measurement of social-emotional wellbeing. A literature search was conducted using the key words: facial masking, hypomimia, individual emotional expressivity/expressive disposition, affective communication, caregiver strain, psychological distress, relationship quality, and quality of life. General and PD-specific

literature were reviewed, where available. As we were focused on the contribution of facial movement to emotional expressivity, literature focusing on other modalities of emotional expression (e.g., speech) were excluded. Item statements were then reviewed as a whole, to check for similarity of content, remove or rework item statements.

Expert Opinion

Experts with experience in working with people with Parkinson's disease were invited to provide their opinions on the preliminary questionnaires. Experts were contacted directly by the first author who explained the goal of research, their rights in providing feedback, and the limits of confidentiality. Experts were sent the questionnaires one to two weeks in advance, before taking part in group or individual discussions about the questionnaire with the researcher. The six experts who took part were registered nurses who work with people who have Parkinson's. Most were experienced in recognizing the signs of FM, but some reported they were less conversant with its functional impact. Familiarity in administering rating scales was highly varied across the expert panel. Experts were asked to provide general opinions of the questionnaires and review each statement providing any responses evoked by the item and suggestions for improvement.

***Content*.** Experts generally thought FMQ content was relevant, appropriate, and comprehensive. Some experts were surprised that SO misread masking as a signal of reduced interest in the relationship or a sign of infidelity. Several items were reworked or refined to make sure statements were unambiguous. A small number of items were removed which experts viewed as too general, redundant, or irrelevant (PwP n = 2, SO n = 3).

Item order. Experts reported the question order was confusing because it jumped between topics, so statements were re-ordered to group similar topics and improve flow. Short statements to demarcate and introduce the topic of each group of statements were added. One expert expressed concern a few questions might be too personal in nature, so potentially sensitive topics were moved towards the end of the checklist, to lessen the likelihood of respondent discomfort. Most experts also liked the mix of positive and negative

question topics, as they thought it would deter respondents becoming indifferent or careless in answering.

***Appearance/Format*.** Experts thought format of the questionnaire made it clear to understand and easy to respond. Tick boxes, rather than circling a response, were added to increase the ease of selecting a response.

Clinical Utility. One expert thought FM did not justify specific assessment, but most judged information obtained by the questionnaire to be useful and relevant. Perceived utility of the questionnaire included: greater confidence to manage FM and related issues, potential to facilitate client learning and motivate clients to assume a role in self-management, normalizing discussion of topics that can be challenging to address (for instance, relationship issues). Because of few existing resources at that time, there was some concern about how to respond to needs for greater FM information/support.

Administration and General Feasibility. Experts cautioned the questionnaires might not be appropriate for clients with little FM knowledge who would require further education to understand and respond to the checklist's questions. Due the personal nature of some question topics, it was also noted rapport would need to be established prior to administration. Another concern was that questionnaire follow-up might impinge on already limited resources. To clarify how to administer the questionnaire, recommendations were added to the FMQ administration and scoring instructions (see Appendix VI). Experts also suggested online/electronic administration, to extend the usability of the questionnaire to a wider range of individuals.

Healthcare Consumer Feedback

Two people with self-reported FM and two partners from the original interviewees provided feedback on the preliminary questionnaires via face-to-face meetings (n = 4). Participants were asked to provide general opinion on the questionnaires, as well feedback on item relevance, comprehensibility, appropriateness, and whether aspects of the FM experience were missing. A small number of items were reworked, refined, or divided using

the consumer feedback.

Content. Minor changes to wording were made to increase the likelihood items were worded plainly and unambiguously. Content was generally viewed as relevant, appropriate, and resonating with personal experience. Rather than being intrusive, participants believed addressing sensitive topics was valuable.

Format. Participants offered positive feedback about format, which was easy to read. The inclusion of open-answer items was also thought to be beneficial because it provided opportunity to express personal views and experiences. However, it was noted some individuals might not be prepared to give an immediate response to open-style questions. Partners valued the opportunity to express their experiences and concerns as some felt no-one had asked what Parkinson's was really like for them.

***Perceived benefits*.** Participants thought the FMQ made it comfortable for people to ask for information or help related to FM. Use of the questionnaires was perceived to be beneficial in three ways: it could help people learn the signs of masking; it helped explain and normalize confusing or strained interpersonal interactions; it may help people start conversations about unspoken or unacknowledged issues. Interviewees also stated the inclusion of direct questions about psychological wellbeing was essential because their mental health had rarely been discussed with healthcare providers in New Zealand.

Administration. It was suggested that some individuals might need to take a break to complete the questionnaire, due to difficulties with concentration and/or fatigue. Although people with Parkinson's estimated the checklist would take them longer to complete than their partners, they did not perceive it to be overly burdensome. Another point of concern was that appropriate and timely follow up is received following administration. To address this issue, guidelines for administration and patient education were added to the FMQ instructions (see Study 2 Appendix VI). Tables 1 and 2 summarize the item content, provenance, and response format for the preliminary versions of the FMQ-PwP and FMQ-SO, respectively.

Table 1.

Item provenance, content, and response format of the FMQ-PwP (preliminary version)

Qualitative Findings	**Quantitative Assessment Instrument**				
Themes, Subthemes, Codes & Interview Quotes	**Domain/ Dimension**	**No.**	**Item**	**Item Response Format**	**Other sources reviewed; then relevant aspects adapted within thematic framework**
Theme 1. Disturbances of Facial Movement and Social-emotional Expressivity **Subtheme 1a**1a "*Watered down*": Muted and Distorted Expressions **Subtheme 1b** The Absence of Expression *"It's shaped different, it's shaped differently and, and it doesn't incorporate the face, you know? The, the… the rest of the face. Yeah it hasn't got all that other subtle stuff that comes with a smile" (P02 – PDparticipant)* *"The majority of the time it's a blank expression…. [laughter]". (P01 – PDparticipant)* *" I noticed when I was talking to people it was almost like I was looking at something behind them. These people would turn to see what I was looking at. But I was looking straight through them" (P08 – PDparticipant)*	FM Severity Estimate	1	**Please rate the overall expression or movement of your face, on everyday basis.**	Closed-ended response (select the best descriptor from four options): *3 =Facial movement and/or expressions are almost always absent.* *2=Facial movement and/or expression are noticeably less intense or less frequent than before Parkinson's developed.* *1=Facial movement and/or expressions appear somewhat less intense or less frequent than before Parkinson's developed.* *0= Facial movement and/or expressions are present and unchanged.*	**Adapted from MDS-UPDRS (item 3.2), & ICRP-PD (items 1, 4, 7)** Reworded to simplify: Expert Opinion
Theme 4. '*It can affect you quite badly without you realising exactly what it is':* Poor Symptom Recognition and Understanding *" I spose in the first instance was when looking at a photograph of myself...That was the first indication that I don't look the same as I used to. Um, I didn't know why. Although I probably thought it had something to do with Parkinson's, but I didn't know whether it did or not. And facial masking I never heard of it, until I read the article in the paper" (P06 – PDparticipant)*	FM Knowledge	2	**Had you heard about the symptom of facial masking (the loss of facial expression or movement) before today?**	Three-point response scale: *I have heard of it and feel informed about it, I have heard of it but do not know much about it, I have not heard of it before*	Reworded to simplify: Researcher Group

Theme 4. '*It can affect you quite badly without you realising exactly what it is'*: Poor Symptom Recognition and Understanding *"Other people notice it, but you don't… So it may affect the way they see things…you may be as happy as them and quite content and think that everything's fine. But when they're looking at it, it's not." (P01 – PDparticipant)*	FM Impact (reversed)	3	**When I am happy people close to me can easily tell what I am feeling.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Berkley Expressivity Questionnaire (BEQ)**
Subtheme 2a. Misattribution of Negative Affect **Theme 4.** '*It can affect you quite badly without you realising exactly what it is':* Poor Symptom Recognition and Understanding *"I feel that people, it's hard, people would misinterpret what I was thinking or how I was reacting to something… and I'd be quite often surprised to find out that someone had said, "oh you know, we just thought you were really grumpy or disengaged or disinterested". (P03 – PDparticipant)*	FM Impact	4	**People comment I appear to be in a bad mood much of the time.** **For e.g., sad, grumpy, bored, cold, disengaged, or uninterested.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Subtheme 1c. Inauthentic and Effortful: Voluntary Expressions "*The smile doesn't come naturally. It's a matter of trying to coordinate a whole lot of muscles… So to smile, you're having to go through a whole thought process as to how am I going to do it. Because it doesn't come naturally"* *(P06 – PDparticipant)*	FM Impact	5	**When I try to make an expression like a smile, it seems to take a lot of effort, feels false, or "put on".**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Code(s): loss of attunement within important relationships *"When I feel very rigid and unable to move and sort of physically stuck, to have that stoicism thrown across your face as well and you're blunted… People can think you have disengaged from your family or from your life and you know, that's really not the case". (P03 – PDparticipant)*	FM Impact (reversed)	6	**My partner shares in my ups and downs.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Family Assessment Device and from MOS- Social Support Survey**
Subtheme 1d. *'I can see him, but I can't':* Facelessness **Code(s):** challenge to self-concept/self-identity *"I don't know who it is. It's not me…. I used to be able to express myself in face and laugh and things like that, I don't know. I look at photographs of me, and I don't think they're me at all…I don't believe it's me" (P09 - PDparticipant).*	FM Impact	7	**It troubles me that my face does not look like me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Subtheme 1d. '*I can see him, but I can't*': Facelessness **Code(s):** Dissonance between experienced and expressed	FM Impact	8	**It troubles me that way I feel inside is**	Four-point dichotomous-ordinal, likert style response scale:	

emotion, bodily autonomy *"It makes me a little bit sad in the fact that it doesn't reflect me. Looking in the mirror doesn't reflect what, how I'm feeling, or what I'm, I should be seeing in the mirror. I'm just seeing someone who's looking at mirror with no expression at all" (P04 - PDparticipant).*			**different from how I look on the outside.**	*Rarely/Never, Sometimes, Often, Almost all the time*	
Subtheme 3a. (reversal) ***'****You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity. **Code(s):** undermining of bond/interaction *"A relationship is built on all those tiny little subtle cues …I know that you understood what I was just talking about by what happened in the corner of your mouth and what happened with your eyes, and so on. You take that away, and nobody thinks oh there's something missing from this relationship but, it's suddenly that trust starts to dissipate. And the communication diminishes and the preparedness to communicate diminishes." (P02 - PDparticipant).*	FM Impact (reversed)	9	**My partner is supportive and understanding when I need to talk or confide in them.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Mos-SSS**
Theme 2. '*If that information's not there, you fill it in':* Misinterpretations of Negative Affect *"I mean [she] might look at me sometimes and, and say 'are you grumpy or something or', and, and when I say no I'm not, I can honestly say no I'm not." (P07 - PDparticipant).*	FM Impact	10	**My partner misreads my emotions, attitude, or communication.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Theme 3. *'Your connections between people are being chipped away at': Social*-emotional Distancing and Disconnection **Code(s):** withdrawal, avoidance **"***It puts…a couple apart. It can… put [Carol] in a, in a place where she doesn't wanna be with me or doesn't wanna to talk to me and I'm in a version of the same. I can't talk to [her], or don't want to talk to [her]" (P04 - PDparticipant).*	FM Impact	11	**I withdraw from people close to me because of my facial appearance.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Facial Disability Index**
Subtheme 1b. The Discomfort of Facial Difference: The Absence of Expression **Code(s):** unpleasant interaction (colleagues & strangers) "*I can remember him saying several times in the middle of meetings you know, 'I can't read you, your face is blank. You never smile' … He said, 'your face would crack if you even tried to smile!*'" (P07 – PDparticipant).	FM Impact	12	**People have been unkind to me because of my facial appearance.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Stigma Scale for Chronic Illnesses**

Theme 1. Unmoving and Unmoved: Disturbances of Facial Movement and Social-emotional Expressivity **Theme 3.** '*Your connections between people are being chipped away at':* Social-emotional Distancing and Disconnection *"Like we're in a dark room or whatever, cause she can't see anything she's got to listen to really what I'm saying… I find it very frustrating…there's something happening that's isn't being passed through by your face or whatever" (P04 - PDparticipant).*	FM Impact (reversed)	13	**My partner can usually tell what I am feeling.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Theme 3. '*Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection **Code(s):** fear negative evaluation of facial appearance by others (public gatherings), social isolation/withdrawal *"I'm inclined to ah, not be, as friendly with people I spose… you sometimes feel that they're looking as though to say 'god he's a gormless wonder now!' So (laughs). That's what I think they're thinking and that alters what I feel, which means I'll drift to the back of whatever's happening." (P07 - PDparticipant).* *"It just got tiring. Yeah I got sick of having to explain the whole time, you know, 'I know I'm not grumpy with you I'm not pissed off I'm just, I just have Parkinson's' yeah. And in the end it was just easier not to go…and that's the trap that you can fall to very easily." (P03- PDparticipant).*	FM Impact	14	**I avoid interactions with others because of what they might think of my face/expression.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Code(s): communication difficulties, feeling unheard, FM & speech difficulty combined *"I get frustrated because I think, you know, she's not listening but the thing is, she never heard what I had to say in the first place, so. And the, when you know, I have a mask on, she can't tell that either even actually said anything or you know" (P03- PDparticipant).*	FM Impact (reversed)	15	**My partner listens to me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Theme 3. '*Your connections between people are being chipped away at':* Social-emotional Distancing and Disconnection *"I think there would've been a distance growing between me and other people because (inaudible) I wasn't showing those subtle signs of empathy….I think possibly I was seen as being unmoved by things". (P02- PDparticipant).*	FM Impact	16	**I feel isolated from people important to me because they can't see emotion on my face.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Facial Disability Index**

Code(s): Compensatory behaviours, extra medication *"So many people came up to me that night and said to me, 'oh my god, [he] looks amazing'… he was getting everyone tequila shots up at the bar, he was on the dance floor! He was just, he was fabulous, he was absolutely fabulous. What people didn't know is he'd doubled his medication and couldn't get out of bed for three days afterwards (voice breaking up)" (P12- Partner).*	FM Impact	17	**I take extra medication to make my face more expressive in social situations.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Reworded to clarify: Expert opinion
[Theme from SO which was included here to test whether it was also true of PwP]	FM Impact	18	**My partner avoids or ignores me because my face does not respond.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Stigma Scale for Chronic Illnesses**
[Theme from SO which was included here to test whether it was also true of PwP]	FM Impact (reversed)	19	**I find my partner's company enjoyable & comfortable**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)**
Code(s): sense of discomfort or unease in close relationships *"I think it's changed the relationship in that, she's tentative. She is tryin' like hell to read me and can't. Even the decision over what we are having for tea, she's sometimes reluctant to make it in case masking shows up as, I don't know if I want that or not. Whereas it probably in all honesty is quite the opposite... So it has affected us, yes" (P07- PDparticipant).*	FM Impact	20	**Interactions with my partner can feel uncomfortable**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Theme 3. *'Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection *"I think [she] …was getting pretty frustrated with my lack of expression… she read into that a distance between us that I wasn't feeling. I think that stuff like becomes self-fulfilling…with that diminishment in subtle signs of caring, I think…self-protection kicks in ….And I think self-protection itself creates distance, and it elicits a response from me....So not trying as much, so distance develops." (P02- PDparticipant).* *"The only thing I've got left is my family and my close friends…It's the only thing left in my life and it's what I'm trying to do... And to have that impacted by…an emotional sort of stonewall um, yeah it's really depressing. It's really hard" (P03- PDparticipant).*	FM Impact	21	**My lack of expression creates a barrier with my partner**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	

NA	Relationship Quality (reverse scored)	22	**I feel loved and wanted by my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from MOSS-SSS**
NA	Relationship Quality	23	**I worry my partner cares less for me than before.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**
NA	Relationship Quality	24	**I think my partner might be better off without me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**
NA	Relationship Quality (reverse scored)	25	**My sexual relationship with my partner is satisfying.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Couples Satisfaction Index (CSI)**
NA	Relationship Quality (reverse scored)	26	**My romantic relationship with my partner is strong and rewarding.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)** Reworded to clarify: Expert Opinion
NA	Relationship Quality	27	**I wish I were closer to my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**
Code: *Frustration*	Relationship Quality	28	**I feel frustrated with my relationship.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
NA	Relationship Quality/	29	**I feel lonely.**	Four-point dichotomous-ordinal, likert style response scale:	**Adapted from Nottingham health Profile**

	Social isolation			*Rarely/Never, Sometimes, Often, Almost all the time*	
NA	Relationship Quality (reverse scored)	30	**I have a close and warm relationship with my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)** Expert consultation – reworded to clarify
NA	Wellbeing Screen, Anxiety	31	**I feel uncertain, worried, or concerned.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from 15 D**
NA	Wellbeing Screen, Stress	32	**I feel angry, irritated, or frustrated.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
NA	Wellbeing Screen, Mood (reversed)	33	**In general, I feel happy or in good spirits.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Reversed to increase balance of content tone: Expert Opinion
Code(s): Compensatory and Coping behaviours	Coping	34	**Please briefly describe any strategies you use to cope with reduced facial expression/ movement. State how helpful, or not helpful, you have found these strategies.**	Open answer	Reworded to simplify: Expert Opinion
Theme 4. '*It can affect you quite badly without you realising exactly what it is*': Poor Symptom Recognition and Understanding	Perceived Overall Impact	35	**Take a moment to reflect on what living with reduced facial expression/movemen t is like for you. Please briefly**	Open answer	Reworded to simplify: Expert Opinion, Researcher Group

			describe your experience here.		
Theme 4. '*It can affect you quite badly without you realising exactly what it is':* Poor Symptom Recognition and Understanding	Perceived Overall Impact	36	**Please rate how the loss of facial expression/movement has affected you and your relationships.**	5-point Likert style: *Not at all, A little, moderately, quite a lot, considerably*	
Subtheme 4a. '*They don't get that information because it's not there and because the harm that's already done in a relationship isn't understood at all'*: Unmet Health Resource Need *"No one's talked to me about the masking thing, at all… if someone had been able to say… There's the facial stuff and that may have been fucking up your relationships for some time… without you realizing it, and people may be not reading you as you think they are. Have a think about that, but here's something to read about that …There should be a hand-out that people get you know. (P02 – PDparticipant)*	FM Health Education Preference	37	**Would you like more information about the topics raised in this questionnaire?**	Dichotomous forced choice: *Yes, Not at the moment*	Reworded to clarify: Researcher group

Table 2.

Item provenance, content, and response format of the FMQ-SO (preliminary version)

Qualitative Findings	**Quantitative Assessment Instrument**				
Themes, Subthemes, Codes & Interview Quotes	**Domain/ Dimension**	**No.**	**Item**	**Item Response Format**	**Other sources reviewed; then relevant aspects adapted within thematic framework**
Theme 1. Unmoving and Unmoved: Disturbances of Facial Movement and Social-emotional Expressivity *"It's interesting, he can be really upset and your face wouldn't tell anything. You know it's that real sort of, dead pan face. You know, he can be saying the words but the face doesn't tell, it doesn't tell anything" (P13 - Partner).* *"Well, a lot of the time he won't do emotions on his face at all. But then sometimes he will really surprise me. He will spontaneously laugh or maybe look really sad or something" (P18 - Partner).*	FM Severity Estimate	1	**Please rate the overall expression or movement of the person with Parkinson's face, on everyday basis.**	Closed-ended response (select the best descriptor from four options): *3 =Facial movement and/or expressions are almost always absent.* *2=Facial movement and/or expression are noticeably less intense or less frequent than before Parkinson's developed.* *1=Facial movement and/or expressions appear somewhat less intense or less frequent than before Parkinson's developed.* *0= Facial movement and/or expressions are present and unchanged.*	**MDS-UPDRS (item 3.2) & ICRP-PD (items 1, 4, 7)** Reworded to simplify: Expert Opinion
Theme 4. '*It can affect you quite badly without you realising exactly what it is'*: Poor Symptom Recognition and Understanding *"It's quite … probably one of the most serious things about the*	FM Knowledge	2	**Had you heard about the symptom of facial masking (the loss of facial expression or**	Three-point response scale: *I have heard of it and feel informed about it, I have heard of it but do not know much about it, I have not heard of it before.*	Reworded to simplify: Researcher Group

Parkinson's … with masking it's a bit subtle, and when you live with someone all of the time, it happens sort of, gradually, and you're not necessarily aware of it. So it can affect you quite badly without you realizing exactly what it is" (P14 – Partner).			**movement) before today?**		
Theme 2. '*If that information's not there, you fill it in':* Misinterpretations of Negative Affect *"if that information's not there, you fill it in and everything you fill in no facial expression with is boredom, tiredness, anger. The really negative emotions" (P12- Partner).*	FM Impact	3	**My partner appears to be in bad mood a lot of the time For e.g. sad, grumpy, bored, cold, disengaged, or uninterested.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Subtheme 3a.' *You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity. *"I can't just look at him and figure out what he thinks 'cause I'm not getting that (laughs) now either. And sometimes I just think I could shake him you know, so…yeah um, I mean we have been married a long time so… used to each other without you know having those um interactions necessarily. But sometimes I just really would like to know (sighs) what he thought about things or you know, (spoken sadly) what he wanted to do or something" (P18 - Partner).*	FM Impact	4	**It is hard for me when my partner's face does not respond.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Berkley Expressivity Questionnaire (BEQ)**
Subtheme 3a. *'You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity. *"It does impact on your enjoyment of life. Because you haven't got someone who's excited when you're excited or sad when you're sad….You haven't got someone you can share that with, at any given time. (Spoken softly) Yeah it does make you sad. Which of course…detracts from your quality of life". (P16 - Partner).*	FM Impact (reversed)	5	**My partner shares in my life's ups and downs.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Code(s): Reduced Displays of positive emotional responsivity *"Cause now he has difficulty smiling so I'm not getting any kind*	FM Impact (reversed)	6	**I can easily tell when my partner is feeling positive emotions.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Berkley Expressivity**

of reaction… Occasionally he will really surprise me and he will really laugh about something, but there is that intermediate thing, you know, with someone just smiling when you say something that they agree with and whatever, that we don't, we don't have that any longer" (P18 - Partner).					**Questionnaire (BEQ)**
Code(s): feeling missed out of expressions *"I love seeing him when he's really animated. I love seeing that side of him again…but if he is with somebody that he doesn't know as well or it's a social occasion and he's trying his best, I feel like they get the absolute best" (P12 - Partner).*	FM Impact	7	**My partner is more expressive with other people, than with me.** For e.g., they try to smile at a social event but not when we are at home alone.	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Theme 2. *'If that information's not there, you fill it in'*: Misinterpretations of Negative Affect **Code(s):** Backtracking, second guessing "I've had to kind of pick myself up because I've looked at [him] and thought, oh my God, he's not interested at all! But then I remember he probably is, but…it doesn't show…I said to him one day, he looks like he's bored shitless! And, and he's not, but that's how the face can sometimes look. You know, and he's not, not really" (P13 - Partner).	FM Impact	8	**I find myself second-guessing what my partner is thinking and feeling.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Reworded to clarify: Expert opinion
Subtheme 2a. Misattribution of Negative Affect **Code(s):** Internally directed negative assumptions about FM *"Sometimes he was looking disapproving … I was being a bit apologetic. And he said to me one day, 'why do you keep apologising? I can't understand why you keep apologising to me all of the time'. And I said, 'because I think you're upset with me in some way'. And he said, 'but I'm not!' "* (P14 - Partner).	FM Impact	9	**I think my partner's unhappiness must be my fault.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
Subtheme 3a. (reversal) '*You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity. **Code(s):** Perceived emotional (un)supportiveness *"It's the not getting the sort of feedback about something that might be quite important… when I try to talk to him…I don't really get any sense of him being particularly worried or caring …but*	FM Impact (reversed)	10	**I can count on my partner to be supportive and understanding.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Family Assessment Device, MOS-Social Support Survey.**

it's those sort of things where maybe I feel like maybe I would like some emotional support over something and (pause, sighs) he doesn't give any indication from his face. I suppose he does more from his actions …It's been more the fact that his face doesn't show emotion I guess". (P18 - Partner).				
Subtheme 2a. Misattribution of Negative Affect **Code(s):** Internally directed negative assumptions about FM *"It can make me feel a bit insecure at times. Um make me feel um, certain rejection sometimes, um make me feel a wee bit upset. Um, a bit oversensitive…. he's at times wondered why I was reacting the way I was…Because I'm trying harder to seek his approval and it was annoying him… I was assuming that he was being disapproving or critical in some way, of me. Mm, judging". (P14 – Partner).*	FM Impact	11	**I feel rejected or criticised when my partner shows little expression.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*
Theme 3. (reversal)*'Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection **Code(s):** perception of indifference, feeling neglected *"Oh it's <u>miserable,</u> it's miserable….Always with him, when you walk into a room and you get home from work and he doesn't seem to even notice that you've walked into the room".* (P12 - Partner).	FM Impact (reversed)	12	**My partner is interested in interacting with me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*
Subtheme 3a. '*You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional Reciprocity. *" I would think did he not hear what I've said, does he not care that this is so exciting or…. I stopped telling him stuff…..not tell him things because I thought he was disapproving or I thought that he was disinterested."* (P10 - Partner).	FM Impact	13	**My partner's lack of expression creates a barrier between us**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*
Code(s): behavioural avoidance, withdrawal *"I don't tend to spend as much time sitting with him like in the evening watching TV. I'm probably more likely to go and do things on the computer or do my um sewing um…I tend to stay at work. I sort of think 'oh, oh nah, I might as well just stay at*	FM Impact	14	**I avoid being near my partner or interacting with my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*

work, I might as well just stay here and potter around and do things here'. I can fill my time in at work. So um, so that is partly because he doesn't respond, I mean there's no, yeah there is no, not so much response in that way". (P16 -Partner).					
Theme 3. (reversal)'*Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection **Subtheme 2a. (reversal)** Misattribution of Negative Affect *"I read into that, that in some way we weren't as close as we were before. It was a perception of something subtle. It wasn't quite the way it was before".* (P14 - Partner).	FM Impact (reversed)	15	**I have a close and warm relationship with my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)**
Subtheme 1d. *'I can see him, but I can't'*: Facelessness *"I've lost…some aspects of that person…the face is definitely one of them. He's still got a lovely face, but...I can't read that face anymore… I can see him, but I can't…It's definitely still him, it's still his lovely face…I just have to really look to see what that face is telling me, you know" (P13 - Partner).* *"It was really difficult because, lots of reasons. One, was I'd look at him and see a different face. I'd see the sternness of his older brother…It was quite disconcerting… And [my husband] is a very humorous and funny person. I don't know this person, you know. I don't know this face".* (P10 - Partner)	FM Impact	16	**It is upsetting my partner's face/expression has changed so much from their former self.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Modified Caregiver Strain Index (CSI)** Reworded to clarify: Expert opinion
Subtheme 2a. Misattribution of Negative Affect **Code(s):** backtracking, compensatory behaviours (challenge cognitions based on FM) *"I have to remind myself all the time that what I think he's feeling is probably not what he's feeling…. I have to remind myself that actually um, he's not, he's not unhappy because of me and the kids...Check in with him a lot...Remind myself of all the good things … just constantly reminding myself ….I struggle with that personally" (P12 - Partner)*	FM Impact	17	**I remind myself how my partner's face looks on the outside probably isn't how they feel inside.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Reworded to clarify: Expert opinion & Consumer feedback
Subtheme 3a. '*You haven't got someone who's excited when you're excited, or sad when you're sad'*: Reduced Emotional	FM Impact (reversed)	18	**My partner's company is**	Four-point dichotomous-ordinal, likert style response scale:	**Adapted from Couples**

Reciprocity. **Code(s):** decreased shared enjoyment/engagement in companionship, sense of unease *"I used to be able to easily tell when he's happy and like with [our hobby] …You'd be enjoying it…and build off that, feed off that.… I don't see that now anymore. You don't get that automatic feedback, and therefore you can't just go with the feel of the moment… We have had to bring in verbal checks…(spoken quietly, looking away & down) which sometimes takes away from the moment, you can't just go with the feel of it"* (P17 - Partner).			**enjoyable and comfortable.**	*Rarely/Never, Sometimes, Often, Almost all the time*	**Satisfaction Index (CSI), Stigma Scale for Chronic Illnesses (SSCI-8)**
Theme 3. '*Your connections between people are being chipped away at':* Social-emotional Distancing and Disconnection **Code(s):** Loneliness, isolation *"His expression doesn't change whether I'm in the room or not … that can be lonely. Just creates this barrier … this distance, it's like this gulf between us or before we've even started any conversation" (P12– Partner)*	FM Impact	19	**I feel disconnected from my partner because their face does not respond to me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Expert consultation – reworded to clarify
Subtheme 2a. Misattribution of Negative Affect **Code(s):** loneliness, questioning commitment or relationship future *"Incredible loneliness for me, like a real sense of um, I don't like where this relationship is going…you know like whoa! This is not who we are, or what, how I communicate, or yeah. It was really quite painful.* (P10 – Partner)	FM Impact	20	**My partner's emotional signals make me concerned they might want to end our relationship.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Expert consultation – reworded to clarify
Subtheme 2a. Misattribution of Negative Affect **Code(s):** undermining trust, questioning fidelity *"I mean we've been together nearly thirty years almo- thir- bloody hell! And I'd never questioned anything in our relationship ever, around loyalty, around fidelity, around trust, anything"* (*P10 – Partner)*	FM Impact	21	**My partner's emotional signals make me concerned they are considering an affair or are having an affair.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Expert consultation – reworded to clarify

Theme 3 (reversal). *'Your connections between people are being chipped away at'*: Social-emotional Distancing and Disconnection ***Code(s):*** loss of emotional relatedness *"I just have to really look to see what that face is telling me, you know...I mean as much as it is hard for me to interpret, it's also hard for him sometimes to share that too… I sure he misses that too, you know….It is part of… your natural life isn't it?"* (P13 - Partner).	FM Impact (reversed)	22	**I feel a strong emotional connection with my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)** Expert consultation – reworded to clarify
Subtheme 1b The Discomfort of Facial Difference: The Absence of Expression *"I think it's affected our sexual relationship…I'll say "don't stare at me like that!" I'm like, ahh, you know! …It's a stare, not an intimate look into your lovers eyes (laughs)…So I find it quite off-putting….I feel really exposed. Invaded almost, you know. Then, well, we laugh. Thank god we can laugh about it, but (pause) it's like, yeah"* (P10 - Partner).	FM Impact	23	**My partner's staring or lack of expression gets in the way of our sexual relationship.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	Expert consultation – reworded to clarify
Subtheme 2a. Misattribution of Negative Affect **Code(s):** Internally directed negative assumptions about FM, self-blame *"I wish I was a lot more oblivious to it because I feel a lot of the time I'm beating myself up… over something that actually um, is probably not even true…. maybe he'd be happier if we weren't around…Maybe he would be happier if I wasn't living at home".* (P12 – Partner)	FM Impact	24	**I think my partner might be better off without me.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**
Subtheme 2a. Misattribution of Negative Affect **Theme 3.** '*Your connections between people are being chipped away at*': Social-emotional Distancing and Disconnection **Code(s):** Internally directed negative assumptions about FM *"Without realising it… well, I have started to make assumptions that the romance is not what it was."* (P14 - Partner).	FM Impact	25	**I worry my partner cares less for me than before.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**

NA	Relationship Quality (reverse scored)	26	**My romantic relationship with my partner is strong and rewarding.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Couples Satisfaction Index (CSI)** Expert consultation to clarify
NA	Relationship Quality	27	**I wish I was closer to my partner.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Experiences in Close Relationships Revised Questionnaire**
Code: *Frustration*	Relationship Quality	28	**I feel frustrated with my relationship.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
NA	Relationship Quality (reverse scored)	29	**My partner makes me feel loved and wanted.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from MOSS-SSS**
Code: *Loneliness*	Relationship Quality / Social Isolation	30	**I feel lonely.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from Nottingham health profile (social isolation)**
NA	Wellbeing Screen, Anxiety	31	**I feel uncertain, worried, or concerned.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	**Adapted from 15 D**
NA	Wellbeing Screen, Stress	32	**I feel angry, irritated, or frustrated.**	Four-point dichotomous-ordinal, likert style response scale: *Rarely/Never, Sometimes, Often, Almost all the time*	
NA	Wellbeing Screen, Mood	33	**In general, I feel happy or in good spirits.**	Four-point dichotomous-ordinal, likert style response scale:	Expert consultation-reversed to

	(reverse scored)			*Rarely/Never, Sometimes, Often, Almost all the time*	increase balance of content tone
Code(s): Compensatory and Coping behaviours	Coping	34	**Please briefly describe any strategies you use to cope with reduced facial expression/ movement. State how helpful, or not helpful, you have found these strategies.**	Open answer	Expert consultation – reworded to simplify
Theme 4. '*It can affect you quite badly without you realising exactly what it is'*: Poor Symptom Recognition and Understanding	Perceived Overall Impact	35	**Take a moment to reflect on what living with reduced facial expression/movement is like for you. Please briefly describe your experience here.**	Open answer	Researcher Group - Expert Consultation – reworded to simplify
Theme 4. '*It can affect you quite badly without you realising exactly what it is'*: Poor Symptom Recognition and Understanding	Perceived Overall Impact	36	**Please rate how the loss of facial expression/movement has affected you and your relationships.**	5-point Likert style: Not at all, A little, moderately, quite a lot, considerably	
Subtheme 4a. '*They don't get that information because it's not there and because the harm that's already done in a relationship isn't understood at all'*: Unmet Health Resource Need *"It's not talked about when they have the meetings…So I don't know what there is really and what you can do to counteract it or that sorta thing"* (P17 - Partner)	FM Health Education Preference	37	**Would you like more information about the topics raised in this questionnaire?**	Dichotomous forced choice Yes, Not at the moment	Researcher group

Study 2 Appendix II. Item-Level Analysis and Item Removal Summary (FMQ-PwP version)

Table 3 shows item completeness and response distributions for each item of the preliminary FMQ-PwP, by self-reported FM status. Table 4 provides item descriptive statistics for the FMQ-PwP items, also by self-reported FM status. Table 5 lists item exclusion criteria and decisions for the FMQ-PwP. Table 6 summarises item reduction of the FMQ-PwP and items which did not fit well psychometrically but were conceptually or clinically important and therefore retained outside the main subscales.

Prior to the main analyses, four items were excluded from the PwP version of the FMQ (#12, #17, #25 & #26). Two items (#25 &26) were excluded for high missing values (greater than or approximately equal to 15% missing data, Tables 3 & 5). Another two items were identified where low response variability was found among respondents reporting FM (#12 & #17, Tables 3 & 5). Item #17 refers to taking extra medication to make one's face more expressive in social situations. Because only 1.9 % (*n* = 1) of respondents with FM said this occurred "sometimes", the item was deemed to be minimally descriptive. Item #12 refers to other people being unkind about one's facial appearance. As only 11.3% of respondents with FM (*n* = 6) answered this occurred "sometimes", this item was also considered unsuitable due to low respondent variability.

Nine items were eliminated during the exploratory factor analysis as they did not contribute to the factor structure (table 6). One item (#23) was removed for loading < .4, and one item (#28) for cross loading (an absolute difference in factor loadings of < .2). Seven items (#13, #10, #3, #16, #24, #29 & #27) were removed for low communalities (< .2).

During reliability analysis of the FMQ-PwP Relationship Difficulty subscale, two items (#19 and #30) showed relatively high corrected item total correlation coefficients and were removed (*r* = .789 and *r* = .756, respectively). One additional item (#6) was deleted to improve reliability. No items were removed from the FMQ-PwP Individual Difficulty Subscale during reliability analysis

Table 3.

Item completeness and response distribution for each FMQ-PwP item, compared by self-reported FM status

	Observed Item Response Distribution									
	Cases reporting FM (*n* = 53)					**Cases NOT reporting FM (*n* = 27)**				
Item	Never or rarely *n* (%)	Sometimes *n* (%)	Often *n* (%)	Almost all the time *n* (%)	Missing *n* (%)	Never or rarely *n* (%)	Sometimes *n* (%)	Often *n* (%)	Almost all the time *n* (%)	Missing *n* (%)
Q3(R)	1 (1.9)	10 (19.2)	13 (25)	28 (53.8)	1 (1.9)	1 (3.8)	3 (11.5)	1 (3.8)	21 (80.8)	1 (3.7)
Q4	27 (50.9)	21 (39.6)	3 (5.7)	2 (3.8)	0 (0)	18 (66.7)	8 (29.6)	1 (3.7)	0 (0.0)	0 (0)
Q5	17 (32.1)	16 (30.2)	12 (22.6)	8 (15.1)	0 (0)	23 (85.2)	4 (14.8)	0 (0.0)	0 (0.0)	0 (0)
Q6 (R)	4 (7.5)	10 (18.9)	16 (30.2)	23 (43.4)	0 (0)	3 (11.5)	8 (30.8)	3 (11.5)	12 (46.2)	1 (3.7)
Q7	18 (34)	25 (47.2)	8 (15.1)	2 (3.8)	0 (0)	23 (85.2)	4 (14.8)	0 (0.0)	0 (0.0)	0 (0)
Q8	18 (34)	20 (37.7)	12 (22.6)	3 (5.7)	0 (0)	17 (63.0)	10 (37.0)	0 (0.0)	0 (0.0)	0 (0)
Q9 (R)	0 (0)	9 (17.0)	5 (9.4)	39 (73.6)	0 (0)	0 (0.0)	3 (11.1)	3 (11.1)	21 (77.8)	0 (0)
Q10	21 (39.6)	25 (17.0)	6 (11.3)	1 (1.9)	0 (0)	11 (42.3)	14 (53.8)	1 (3.8)	0 (0.0)	1 (3.7)
Q11	36 (67.9)	13 (24.5)	3 (5.7)	1 (1.9)	0 (0)	26 (96.3)	1 (3.7)	0 (0.0)	0 (0.0)	0 (0)
Q12	47 (88.7)	6 (11.3)	0 (0)	0 (0)	0 (0)	2 (96.3)	1 (3.7)	0 (0.0)	0 (0.0)	0 (0)
Q13 (R)	3 (5.7)	15 (28.3)	19 (35.8)	16 (30.2)	0 (0)	2 (7.7)	6 (23.1)	4 (15.4)	14 (53.8)	1 (3.7)
Q14	35 (66)	14 (26.4)	4 (7.5)	0 (0)	0 (0)	27 (100)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0)
Q15 (R)	1 (2.0)	8 (15.7)	8 (15.7)	34 (66.7)	2 (3.9)	1 (3.7)	3(11.1)	8 (29.6)	15 (55.6)	0 (0)
Q16	37 (71.2)	12 (23.1)	2 (3.8)	1 (1.9)	1 (1.9)	25 (96.2)	1 (3.8)	0 (0.0)	0 (0.0)	1 (3.7)
Q17	51 (98.1)	1 (1.9)	0 (0)	0 (0)	1 (1.9)	27 (100)	0 (0.0)	0 (0.0)	0 (0.0)	0 (0)
Q18	48 (90.6)	4 (7.5)	1 (1.9)	0 (0)	0 (0)	26 (100)	0 (0.0)	0 (0.0)	0 (0.0)	1 (3.7)
Q19 (R)	0 (0)	3 (5.7)	8 (15.1)	41 (77.4)	0 (0)	1 (3.8)	1 (3.8)	1 (3.8)	24 (92.3)	1 (3.7)
Q20	37 (69.8)	14 (26.4)	2 (3.8)	0 (0)	0 (0)	21 (80.8)	4 (15.4)	1 (3.8)	0 (0.0)	1 (3.7)
Q21	41 (77.4)	10 (18.9)	2 (3.8)	0 (0)	0 (0)	24 (92.3)	2 (7.7)	0 (0.0)	0 (0.0)	1 (3.7)
Q22 (R)	0 (0)	2 (3.8)	5 (9.4)	43 (81.1)	0 (0)	1 (3.8)	2 (7.7)	1 (3.8)	22 (84.6)	1 (3.7)
Q23	39 (78)	9 (17.0)	1 (1.9)	1 (1.9)	0 (0)	22 (88.0)	3 (12.0)	0 (0.0)	0 (0.0)	2 (7.4)
Q24	34 (69.4)	9 (17.0)	5 (9.4)	1 (1.9)	0 (0)	18 (69.2)	8 (30.8)	0 (0.0)	0 (0.0)	1 (3.7)
Q25 (R)	8 (20.5)	8 (15.1)	8 (15.1)	15 (28.3)	14 (26.4)	3 (37.5)	0 (0.0)	1 (12.5)	4 (50.0)	19 (70.4)
Q26 (R)	2 (4.2)	8 (15.1)	12 (22.6)	26 (49.1)	5 (9.4)	1 (5.6)	2 (11.1)	5 (27.8)	10 (55.6)	9 (33.3)
Q27	24 (49)	15 (28.3)	6 (11.3)	4 (7.5)	4 (7.5)	14 (66.7)	5 (23.8)	1 (4.8)	1 (4.8)	6 (22.2)
Q28	31 (62)	15 (28.3)	4 (7.5)	0 (0)	3 (5.7)	16 (69.6)	7 (30.4)	0 (0.0)	0 (0.0)	4 (14)
Q29	27 (50.9)	21 (39.6)	3 (5.7)	2 (3.8)	0 (0)	13 (50.0)	8 (30.8)	5 (19.2)	0 (0.0)	1 (3.7)
Q30 (R)	0 (0)	4 (7.5)	6 (11.3)	40 (75.5)	3 (5.7)	1 (4.5)	2 (9.1)	2 (9.1)	17 (77.3)	5 (18.5)

Table 4

Descriptive statistics of the FMQ-PwP, compared by self-reported FM status

Descriptive Statistics																
	Cases reporting FM (*n* = 53)								Cases NOT reporting FM (*n* = 27)							
Item	**Mean**	**Median**	**Mode**	**SD**	**Min**	**Max**	**Skew**	**Kurtosis**	**Mean**	**Median**	**Mode**	**SD**	**Min**	**Max**	**Skew**	**Kurtosis**
Q3(R)	0.69	0.0	0.0	0.85	0	3	0.85	-0.51	0.38	0.0	0.0	0.85	0	3	2.07	3.15
Q4	0.62	0.0	0.0	0.77	0	3	1.31	1.80	0.37	0.0	0.0	0.56	0	2	1.25	0.74
Q5	1.21	1.0	0.0	1.06	0	3	0.37	-1.09	0.15	0.0	0.0	0.36	0	1	2.10	2.59
Q6(R)	0.91	1.0	0.0	0.97	0	3	0.73	-0.54	1.08	1.0	0.0	1.13	0	3	0.38	-1.42
Q7	0.89	1.0	1.0	0.80	0	3	0.68	0.15	0.15	0.0	0.0	0.36	0	1	2.10	2.59
Q8	1.00	1.0	1.0	0.90	0	3	0.50	-0.60	0.37	0.0	0.0	0.49	0	1	0.57	-1.82
Q9(R)	0.43	0.0	0.0	0.77	0	2	1.40	0.22	0.33	0.0	0.0	0.68	0	2	1.85	2.08
Q10	0.75	1.0	1.0	0.73	0	3	0.73	0.35	0.65	1.0	1.0	0.69	0	3	1.38	3.92
Q11	0.42	0.0	0.0	0.69	0	3	1.77	3.06	0.04	0.0	0.0	0.19	0	1	5.20	27.00
Q12	0.11	0.0	0.0	0.32	0	1	2.51	4.48	0.04	0.0	0.0	0.19	0	1	5.20	27.00
Q13(R)	1.09	1.0	1.0	0.90	0	3	0.30	-0.84	0.85	0.0	0.0	1.05	0	3	0.78	-0.82
Q14	0.42	0.0	0.0	0.63	0	2	1.28	0.56	0.00	0.0	0.0	0.00	0	0	-	-
Q15(R)	0.53	0.0	0.0	0.83	0	3	1.31	0.45	0.63	0.0	0.0	0.84	0	3	1.25	0.99
Q16	0.37	0.0	0.0	0.66	0	3	2.03	4.49	0.12	0.0	0.0	0.59	0	3	5.10	26.00
Q17	0.02	0.0	0.0	0.14	0	1	7.21	52.00	0.00	0.0	0.0	0.00	0	0	-	-
Q18	0.11	0.0	0.0	0.38	0	2	3.57	13.32	0.00	0.0	0.0	0.00	0	0	-	-
Q19(R)	0.27	0.0	0.0	0.56	0	2	2.03	3.22	0.22	0.0	0.0	0.70	0	3	3.33	10.90
Q20	0.34	0.0	0.0	0.55	0	2	1.39	1.06	0.27	0.0	0.0	0.67	0	3	3.14	11.23
Q21	0.26	0.0	0.0	0.52	0	2	1.90	2.89	0.08	0.0	0.0	0.27	0	1	3.37	10.16
Q22(R)	0.18	0.0	0.0	0.48	0	2	2.77	7.18	0.31	0.0	0.0	0.79	0	3	2.56	5.74
Q23	0.28	0.0	0.0	0.61	0	3	2.63	8.01	0.12	0.0	0.0	0.33	0	1	2.49	4.56
Q24	0.45	0.0	0.0	0.77	0	3	1.63	1.86	0.31	0.0	0.0	0.47	0	1	0.89	-1.32
Q25(R)	1.23	1.0	0.0	1.18	0	3	0.34	-1.41	1.25	0.5	0.0	1.49	0	3	0.48	-2.25
Q26(R)	0.71	0.0	0.0	0.90	0	3	0.99	-0.08	0.67	0.0	0.0	0.91	0	3	1.30	1.08
Q27	0.80	1.0	0.0	0.96	0	3	1.02	0.09	0.48	0.0	0.0	0.81	0	3	1.93	3.75
Q28	0.46	0.0	0.0	0.65	0	2	1.10	0.15	0.30	0.0	0.0	0.47	0	1	0.91	-1.29
Q29	0.62	0.0	0.0	0.77	0	3	1.31	1.80	0.69	0.5	0.0	0.79	0	2	0.63	-1.07
Q30(R)	0.28	0.0	0.0	0.61	0	2	2.06	3.05	0.41	0.0	0.0	0.85	0	3	2.08	3.50

Table 5

Item Screening of the FMQ-PwP, by self-reported FM status

	Item Exclusion Criteria											
	Cases Reporting FM (*n* = 53)						All cases (*N* = 80)					
Item	I. Mean close to mid score (±1)	II. Mean & median similar (± 0.50)	III. SD >0.75	IV. Responses used ≥3	Failed (I-IV)	Decision *	I. Mean close to mid score (±1)	II. Mean & median similar (± 0.50)	III. SD >0.75	IV. Responses used ≥3	Failed (I-IV)	Decision *
Q3(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q4	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	No	Yes	1	RETAIN
Q5	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q6(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q7	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q8	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q9(R)	No	Yes	Yes	No	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q10	Yes	Yes	No	Yes	1	RETAIN	Yes	Yes	No	Yes	1	RETAIN
Q11	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q12	No	Yes	No	No	3	**REMOVE**	No	Yes	No	No	3	**REMOVE**
Q13(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q14	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q15(R)	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	1	RETAIN
Q16	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q17	No	Yes	No	No	3	**REMOVE**	No	Yes	No	No	3	**REMOVE**
Q18	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q19(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q20	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q21	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q22(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q23	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q24	No	Yes	Yes	Yes	1	RETAIN	No	Yes	No	Yes	1	RETAIN
Q25(R)	Yes	Yes	Yes	Yes	0	**REMOVE**	Yes	Yes	Yes	Yes	0	**REMOVE**
Q26(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	No	Yes	Yes	1	**REMOVE**
Q27	Yes	Yes	Yes	Yes	0	RETAIN	Yes	No	Yes	Yes	1	RETAIN
Q28	No	Yes	No	Yes	1	RETAIN	No	Yes	No	Yes	2	RETAIN
Q29	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	1	RETAIN
Q30(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN

Note. * Remove if ≥3 criteria failed. Items #25 and #26 removed for missing values >15%

Table 6

Summary of item removal decisions, during development and testing of the FMQ-PwP

Item	Abbreviated Item Content	Stage at Item Removal	Item Evaluation	Retained outside main scales*	Rationale *
Q17	Extra medication to improve expression	Item Acceptability	Low Response Variability	**Yes**	Clinically useful to identify if medicating safely.
Q12	Others have been unkind about face		Low Response Variability	**Yes**	Rare but conceptually important to some individuals with FM.
Q25	General relationship quality	Suitability of Data for EFA	High proportion missing values (>15%)	No	
Q26	Intimacy quality		High proportion missing values (>15%)	**Yes**	Sensitive topic relevant to some individuals with FM.
Q28	Frustration with relationship	Factorial Structure of Questionnaire	Cross loading	No	
Q23	Worry partner cares less		Low loading (<.4)	No	
Q3	Others can see if feeling happy		Low communality (<.2)	**Yes**	Clinically relevant in assessing symptom awareness.
Q10	Partner misreads mental state		Low communality (<.2)	**Yes**	Clinically relevant in evaluating insight of FM impact.
Q13	Partner can read mental state		Low communality (<.2)	**Yes**	Clinically relevant in evaluating insight of FM impact.
Q16	Feel isolated as cannot show others emotion		Low communality (<.2)	**Yes**	Rare yet conceptually important to some individuals with FM.
Q24	Partner better off without me		Low communality (<.2)	No	
Q29	Loneliness		Low communality (<.2)	No	
Q27	Closeness with partner		Low communality (<.2)	No	
Q6	Partner shares in ups and downs	Internal Consistency Analysis	Improve Reliability	No	
Q19	Enjoyment of companionship		Item Redundancy	No	-
Q30	General relationship quality		Item Redundancy	No	-

Note. * Items excluded from the main analyses but retained in the FMQ total scale. Such items reflected conceptually or clinically important topics but did not behave as expected in the psychometric analyses. These items are worthy of further investigation.

Study 2 Appendix III: Item-Level Analysis and Item Removal Summary (FMQ-SO version)

Table 7 shows item completeness and response distributions for each item of the preliminary FMQ-SO, by SO-reported FM status. Table 8 provides item descriptive statistics for the FMQ-SO items, also by SO-reported FM status. Table 9 lists item exclusion criteria and decisions for the FMQ-SO. Table 10 summarises item reduction of the FMQ-SO and items which did not fit well psychometrically but were conceptually or clinically important and therefore retained outside the main subscales.

Six items were excluded from SO version of the FMQ prior to the main analyses (#14, #20, #21, #23 & #26, tables 7 & 8). Two items (#23 & #26) were excluded for high missing values (≥ 15% missing data). Three items were identified as having low response variability among FM-reporting respondents (# 14, #20 & #21). Item #14 refers avoiding being near one's partner, with 20.4% reporting this occurred "sometimes" (*n* = 10). Item #21 refers to concern one's partner might be having an affair because of the emotional signals they appear to display. Additionally, on item #20 respondents with FM used all response categories, but the three highest value options were endorsed by just one respondent each. Item #20 was conceptually similar to item #21, as it refers to concern one's partner might want to end the relationship because of the emotional signals they appear to display.

During screening for suitability of data for factor analysis, one item (#7) was removed based on a lack of bivariate correlations > .2. No further items were eliminated from the FMQ-SO during the exploratory factor analysis. In the internal reliability analysis, three items (#13, #18, #19) were identified with patterns of comparatively high inter-item correlations (> .7) with at least four other items, and corrected item-totals relatively close to one (*r* = .828, *r* = .852, and *r* = .814, respectively). The largest inter-item correlation was observed between #13 and #19 (*r* = .782). On examination, the conceptual content of item #13 item was similar to item 19. In addition, a second cluster of inter-item correlations was observed between items #27, #28 & #30. These three items also had somewhat elevated corrected item-totals

(*r* = .722, *r* = .712, and *r* = .719, respectively). Given this, it was decided to remove items #13, #18, #28 and #30.

Table 7

Item completeness and response distribution for each FMQ -SO item, compared by self-reported FM status

	Observed Item Response Distribution									
	Cases reporting FM (n = 50)					**Cases NOT reporting FM *(n = 8)***				
Item	Never or rarely *n* (%)	Sometimes *n* (%)	Often *n* (%)	Almost all the time *n* (%)	Missing *n* (%)	Never or rarely *n* (%)	Sometimes *n* (%)	Often *n* (%)	Almost all the time *n* (%)	Missing *n* (%)
Q3	10 (20.83)	29 (60.4)	6 (12.0)	3 (6.0)	2 (4.0)	4 (50)	4 (50)	0 (0)	0 (0)	0 (0)
Q4	11 (22.9)	28 (58.3)	7 (14.6)	2 (4.2)	2 (4.0)	7 (87.5)	1 (12.5)	0 (0)	0 (0)	0 (0)
Q5 (R)	5 (10.2)	11 (22.4)	14 (28.6)	19 (38.8)	1 (2.0)	1 (12.5)	0 (0)	2 (25.0)	5 (62.5)	0 (0)
Q6 (R)	3 (6.3)	13 (27.21)	14 (29.2)	18 (37.5)	2 (4.0)	0 (0)	1 (12.5)	1 (12.5)	5 (62.5)	0 (0)
Q7	14 (28.6)	20 (40.8)	14 (28.6)	1 (2.0)	1 (2.0)	6 (75.0)	2 (25.0)	0 (0)	0 (0)	0 (0)
Q8	6 (12.5)	29 (60.4)	9 (18.8)	4 (8.3)	2 (4.0)	3 (37.5)	5 (62.5)	0 (0)	0 (0)	0 (0)
Q9	30 (61.2)	16 (32.7)	2 (4.1)	1 (2.0)	1 (2.0)	7 (87.5)	1 (12.5)	0 (0)	0 (0)	0 (0)
Q10 (R)	1 (2.0)	14 (28.0)	7 (14.0)	28 (56.0)	0 (0)	0 (0)	0 (0)	1 (12.5)	7 (87.5)	0 (0)
Q11	27 (54.0)	19 (38.0)	4(8.0)	0 (0)	0 (0)	7 (87.5)	1 (12.5)	0 (0)	0 (0)	0 (0)
Q12 (R)	1 (2.0)	11 (22.0)	18 (36.0)	20 (40.0)	0 (0)	0 (0)	1 (12.5)	2 (25.0)	5 (62.5)	0 (0)
Q13	21 (43.8)	21 (43.8)	5 (10.4)	1 (2.1)	2 (4.0)	6 (75.0)	1 (12.5)	0 (0)	0 (0)	0 (0)
Q14	39 (79.6)	10 (20.4)	0 (0)	0 (0)	1 (2.0)	8 (100)	0 (0)	0 (0)	0 (0)	0 (0)
Q15 (R)	3 (6.1)	3 (6.1)	4 (8.2)	39 (79.6)	1 (2.0)	0 (0)	0 (0)	0 (0)	8 (100)	0 (0)
Q16	14 (29.2)	14 (29.2)	11 (22.9)	9 (18.8)	2 (4.0)	5 (62.5)	1 (12.5)	0 (0)	0 (0)	0 (0)
Q17	14 (28.6)	19 (38.8)	11 (22.4)	5 (10.2)	1 (2.0)	6 (75.0)	1 (12.5)	0 (0)	0 (0)	1 (12.5)
Q18 (R)	1 (2.0)	12 (24.5)	9 (18.4)	27 (55.1)	1 (2.0)	0 (0)	0 (0)	1 (12.5)	7 (87.5)	0 (0)
Q19	25 (52.1)	16 (33.3)	6 (12.5)	1 (2.1)	2 (4.0)	7 (87.5)	0 (0)	0 (0)	0 (0)	1 (12.5)
Q20	44 (93.6)	1 (2.1)	1 (2.1)	1 (2.1)	3 (6.0)	7 (87.5)	0 (0)	0 (0)	0 (0)	1 (12.5)
Q21	46 (97.9)	1 (2.1)	0 (0)	0 (0)	0 (0)	8 (100)	0 (0)	0 (0)	0 (0)	0 (0)
Q22 (R)	2 (4.1)	4 (8.2)	3 (6.1)	40 (81.6)	1 (2.0)	8 (100)	0 (0)	0 (0)	0 (0)	0 (0)
Q23	29 (70.7)	5 (12.2)	4 (9.8)	3 (7.3)	9(18.0)	4 (50.0)	0 (0)	0 (0)	0 (0)	4 (50.0)
Q24	44 (91.7)	3 (6.3)	0 (0)	1 (2.1)	2 (4.0)	6 (75.0)	0 (0)	0 (0)	0 (0)	2 (25.0)
Q25	40 (83.3)	6 (12.5)	2 (4.2)	0 (0)	2 (4.0)	6 (75.0)	0 (0)	0 (0)	0 (0)	2 (25.0)
Q26 (R)	4 (9.1)	11 (25.0)	10 (22.7)	19 (43.2)	6 (12)	0 (0)	0 (0)	0 (0)	6 (75.0)	2 (25.0)
Q27	18 (38.3)	18 (38.3)	8 (17.0)	3 (6.4)	3 (6.0)	5 (62.5)	0 (0)	0 (0)	0 (0)	3 (37.5)
Q28	24 (49.0)	20 (40.8)	4 (8.2)	1 (2.0)	1 (2.0)	4 (50)	1 (12.5)	0 (0)	0 (0)	3 (37.5)
Q29 (R)	4 (8.2)	9 (18.4)	12 (24.5)	24 (49.0)	1 (2.0)	0 (0)	0 (0)	2 (25.0)	6 (75.0)	0 (0)
Q30	24 (50.0)	20 (41.7)	2 (4.2)	2 (4.2)	2 (4.0)	7 (87.5)	0 (0)	0 (0)	0 (0)	1 (12.5)

Table 8

Descriptive statistics of the FMQ-SO, compared by self-reported FM status

Descriptive Statistics																
	Cases reporting FM (*n* = 50)								Cases NOT reporting FM (*n* = 8)							
Item	**Mean**	**Median**	**Mode**	**SD**	**Min**	**Max**	**Skew**	**Kurtosis**	**Mean**	**Median**	**Mode**	**SD**	**Min**	**Max**	**Skew**	**Kurtosis**
Q3(R)	1.04	1.00	1	.771	0	3	.801	.976	.50	.50	0	.535	0	1	.000	-2.800
Q4	1.00	1.00	1	.744	0	3	.648	.748	.13	.00	0	.354	0	1	2.828	8.000
Q5	2.00	2.00	2	.744	0	3	-.648	.748	.63	.00	0	1.061	0	3	1.960	3.937
Q6(R)	1.04	1.00	0	1.020	0	3	.530	-.899	.75	.00	0	1.165	0	3	1.355	.620
Q7	1.02	1.00	0	.956	0	3	.414	-.960	.25	.00	0	.463	0	1	1.440	.000
Q8	1.04	1.00	1	.815	0	3	.164	-.901	.63	1.00	1	.518	0	1	-.644	-2.240
Q9(R)	1.23	1.00	1	.778	0	3	.699	.505	.13	.00	0	.354	0	1	2.828	8.000
Q10	.47	.00	0	.680	0	3	1.565	2.839	.13	.00	0	.354	0	1	2.828	8.000
Q11	.76	.00	0	.938	0	3	.663	-1.178	.13	.00	0	.354	0	1	2.828	8.000
Q12	.54	.00	0	.646	0	2	.794	-.361	.63	.00	0	1.061	0	3	1.960	3.937
Q13(R)	.86	1.00	0	.833	0	3	.493	-.776	.14	.00	0	.378	0	1	2.646	7.000
Q14	.71	1.00	0	.743	0	3	.860	.530	.00	.00	0	-	0	0	-	-
Q15 (R)	.20	.00	0	.407	0	1	1.515	.307	.00	.00	0	-	0	0	-	-
Q16	.39	.00	0	.862	0	3	2.197	3.718	.17	.00	0	.408	0	1	2.449	6.000
Q17	1.31	1.00	0	1.095	0	3	.250	-1.229	.14	.00	0	.378	0	1	2.646	7.000
Q18	1.14	1.00	1	.957	0	3	.445	-.688	.13	.00	0	.354	0	1	2.828	8.000
Q19(R)	.73	.00	0	.908	0	3	.737	-.937	.00	.00	0	-	0	0	-	-
Q20	.65	.00	0	.785	0	3	1.005	.327	.00	.00	0	-	0	0	-	-
Q21	.13	.00	0	.536	0	3	4.537	21.018	.00	.00	0	-	0	0	-	-
Q22(R)	.02	.00	0	.146	0	1	6.856	47.000	.00	.00	0	-	0	0	-	-
Q23	.35	.00	0	.805	0	3	2.273	4.118	.00	.00	0	-	0	0	-	-
Q24	.54	.00	0	.951	0	3	1.630	1.425	.00	.00	0	-	0	0	-	-
Q25(R)	.13	.00	0	.489	0	3	4.870	26.373	.00	.00	0	-	0	0	-	-
Q26(R)	.21	.00	0	.504	0	2	2.455	5.463	.00	.00	0	-	0	0	-	-
Q27	1.00	1.00	0	1.034	0	3	.528	-1.025	.00	.00	0	-	0	0	-	-
Q28	.91	1.00	0	.905	0	3	.725	-.234	.20	.00	0	.447	0	1	2.236	5.000
Q29	.63	1.00	0	.727	0	3	1.039	.989	.25	.00	0	.463	0	1	1.440	.000
Q30(R)	.86	1.00	0	1.000	0	3	.819	-.537	.00	.00	0	.000	0	0	-	-

Table 9

Item Screening of the FMQ-SO, by self-reported FM status

	Item Exclusion Criteria											
	Cases Reporting FM (*n* = 50)						All cases (*N* = 58)					
Q3(R)	I. Mean close to mid score (±1)	II. Mean & median similar (± 0.50)	III. SD >0.75	IV. Responses used ≥3	Failed (I-IV)	Decision *	I. Mean close to mid score (±1)	II. Mean & median similar (± 0.50)	III. SD >0.75	IV. Responses used ≥3	Failed (I-IV)	Decision *
Q4	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q5	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	1	RETAIN
Q6(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q7	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q8	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q9(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q10	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q11	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	1	RETAIN
Q12	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q13(R)	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	3	**REMOVE**
Q14	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	0	RETAIN
Q15 (R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q16	No	Yes	Yes	Yes	1	RETAIN	No	Yes	Yes	Yes	1	RETAIN
Q17	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	2	RETAIN
Q18	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	3	**REMOVE**
Q19(R)	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	2	RETAIN
Q20	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	2	RETAIN
Q21	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q22(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q23	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	2	RETAIN
Q24	No	Yes	Yes	Yes	1	**REMOVE**	No	Yes	Yes	Yes	2	RETAIN
Q25(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	1	RETAIN
Q26(R)	No	Yes	No	Yes	2	RETAIN	No	Yes	No	Yes	0	**REMOVE**
Q27	Yes	Yes	Yes	Yes	0	**REMOVE**	Yes	Yes	Yes	Yes	1	**REMOVE**
Q28	Yes	Yes	Yes	Yes	0	RETAIN	Yes	Yes	Yes	Yes	1	RETAIN
Q29	Yes	No	No	Yes	2	RETAIN	Yes	No	No	Yes	2	RETAIN
Q30(R)	Yes	No	Yes	Yes	1	RETAIN	Yes	No	Yes	Yes	1	RETAIN
Q3(R)	Yes	No	No	Yes	2	RETAIN	Yes	No	No	Yes	2	RETAIN

Note. * Remove if ≥3 criteria failed. Items #23 and #26 removed for missing values >15%

Table 10

Summary of item removal decisions, during development and testing of the FMQ-SO

Item	Abbreviated Item Content	Stage at Item Removal	Item Evaluation	Retained outside main scales*	Rationale *
Q14	Avoids partner	Item Evaluation	Low Response Variability	**Yes**	Rare but conceptually important
Q20	Concerns about relationship strength		Low Response Variability	**Yes**	Rare but conceptually important
Q21	Concerns partner unfaithful		Low Response Variability	**Yes**	Rare but conceptually important
Q23	Staring is off-putting (physical intimacy)	Suitability Screening for EFA	High proportion missing values (>15%)	**Yes**	Sensitive topic relevant to some individuals.
Q26	Emotional Intimacy quality		High proportion missing values (>15%)	**Yes**	Sensitive topic relevant to some individuals.
Q7	Partner more expressive with others		Low inter-item correlations (few >.3)	**Yes**	Rare but conceptually important
Q13	Lack of expression creates a barrier	Internal Consistency Analysis	Improve Reliability	No	
Q18	Enjoyment of companionship		Improve Reliability	No	
Q28	Frustration with relationship		Item Redundancy	No	
Q30	Loneliness		Item Redundancy	No	

Note. *Items excluded from the main analyses but retained in the FMQ total scale. Such items reflected conceptually or clinically important topics but did not behave as expected in the psychometric analyses. These items are worthy of further investigation.

Study 2 Appendix IV: Principle Axis factoring of the FMQ

FMQ-PwP

A principle axis factor (PAF) analysis was conducted with orthogonal (varimax) rotation for the FMQ-PwP, as initial analyses revealed factors were not correlated. Table 11 shows a comparative summary of the factor structure of the FMQ-PwP, before and after removal of items as a result of the reliability analysis.

Initial Factor Matrix. KMO measure of sampling adequacy (*KMO* = 0.731) and Bartlett's test of Sphericity ($p < 0.001$) were acceptable. Scree plot inspection and Eigenvalues of >1 identified a two-factor solution with eigenvalues of 4.56 and 3.24 (factor 1 and 2, respectively), explaining 45.55% of the total variance.

Final Factor Matrix (following removal of items to reduce redundancy and improve reliability). KMO measure of sampling adequacy (*KMO* = 0.71) and Bartlett's test of Sphericity ($p < 0.001$) met predetermined criteria. A two-factor solution was retained with eigenvalues of 3.70 and 2.61 (factor 1, factor 2 respectively), explaining 44.03% of the total variance. The first and second factors switched after the removal of items to reduce redundancy and improve reliability.

FMQ-SO

A principle axis factor (PAF) analysis was conducted for the FMQ-SO. Table 12 shows a comparative summary of the factor structure of the FMQ-SO, before and after removal of items as a result of the reliability analysis.

Initial Factor Matrix*.* KMO measure of sampling adequacy (*KMO* = 0.852) and Bartlett's test of Sphericity ($p < 0.001$) were adequate. Scree plot inspection and Eigenvalues of > 1 identified a single factor solution as the most clinically interpretable with an eigenvalue of 10.39, explaining 45.19% of the total variance.

Final Factor Matrix (following removal of items to reduce redundancy and improve reliability). KMO measure of sampling adequacy (*KMO* = 0.842) and Bartlett's test of

Sphericity ($p < 0.001$) were acceptable. A single factor solution was retained with an eigenvalue of 7.50, explaining 41.67% of the total variance.

Table 11

Comparative summary showing the factor matrix of the FMQ-PwP, before and after removal of items during reliability analysis

Initial						Final*					
Subscale	**Item**	**Factor 1**	**Factor 2**	**Variance**	**α**	**Subscale**	**Item**	**Factor 1**	**Factor 2**	**Variance**	**α**
FM-related Relation-ship Difficulty	Q19 (R)	**0.834**	.080	26.98%	.850	FM-related Individual Difficulty	Q 8	**.845**	-.086	26.7%	.840
	Q30 (R)	**0.782**	.119				Q7	**.787**	-.057		
	Q15 (R)	**0.727**	.062				Q11	**.726**	.126		
	Q9 (R)	**0.710**	-.072				Q14	**.680**	.147		
	Q22 (R)	**0.630**	-.011				Q5	**.657**	.099		
	Q6 (R)	**0.525**	-.076				Q4	**.425**	.189		
	Q18	**0.507**	.186			FM-related Relation-ship Difficulty	Q15 (R)	.036	**.791**	17.3%	.770
	Q21	**0.477**	.196				Q9 (R)	-.070	**.643**		
	Q20	**0.466**	.113				Q22 (R)	-.026	**.589**		
FM-related Individual Difficulty	Q8	-.093	**0.846**	18.56%	.830		Q21	.177	**.549**		
	Q7	-.035	**0.776**				Q18	.165	**.540**		
	Q11	.095	**0.727**				Q20	.109	**.443**		
	Q14	.149	**0.683**								
	Q5	.070	**0.660**								
	Q4	-.093	**0.429**								

Note. Two factor solution retained using Principle Axis Factoring and Varimax Rotation.
* EFA rerun after the removal of items to reduce redundancy and improve reliability.

Table 12

Comparative summary showing the factor matrix of the FMQ-SO, before and after items were removed due to results of the reliability analysis

		Initial					Final *		
Scale	**Item**	**Factor 1**	**Variance**	**α**	**Scale**	**Item**	**Factor 1**	**Variance**	**α**
FM Difficulty	Q18(R)	.807	45.2%	.953	FM Difficulty	Q19	.794	41.7 %	.936
	Q19	.804				Q29(R)	.742		
	Q13	.791				Q3	.737		
	Q4	.744				Q10(R)	.737		
	Q8	.739				Q8	.733		
	Q30	.735				Q4	.729		
	Q28	.733				Q27	.683		
	Q10(R)	.729				Q16	.676		
	Q29(R)	.719				Q11	.671		
	Q3	.717				Q15(R)	.653		
	Q27	.712				Q24	.624		
	Q16	.690				Q5(R)	.570		
	Q11	.677				Q22(R)	.566		
	Q15(R)	.636				Q12(R)	.546		
	Q24	.633				Q6(R)	.540		
	Q22(R)	.561				Q9	.525		
	Q6(R)	.556				Q17	.495		
	Q12	.534				Q25	.469		
	Q5(R)	.532							
	Q9	.526							
	Q17	.491							
	Q25	.485							

Note. Single factor solution with Principle Axis Factoring.
* EFA rerun after the removal of items to reduce redundancy and improve reliability.

Study 2 Appendix V. Suggestions for future revisions

The final questionnaires and instructions for scoring and interpretation are presented in Appendix VI. Further development of the questionnaires could focus on the following recommendations.

Item Response Options. Only a small proportion of respondents endorsed the highest value on items of both versions of the FMQ. Typically, those respondents were individuals who described the highest severity of FM. Based on this finding, the FMQ response options could be modified to aggregate the two highest response options (*"*often" and "*almost all of the time")* into one "*often*" option, and the middle option expanded to "*occasionall*y" and "*somewhat often"*.

To make it easier for answer for individuals who have FM but experience few associated problems, or who attribute the cause of a statement to other symptoms of PD (such as speech difficulties), we suggest adding a "*not applicable*" or "*because of another aspect of PD"* option to the response scale.

Scale redundancy / Capturing different aspects of FM-related difficulty. Internal consistency analyses indicated there may be some redundancy in the FMQ-PwP and FMQ-SO. In general, future researchers may wish to reword some items to reduce ambiguity and better represent thematic content of the experiences of people with FM in PD. On the FMQ-SO this might include the following: "*My partner is interested in interacting with me*" could be reworded as "*It feels like my partner does not notice me*"; and "*My partner's company is enjoyable and comfortable"* could be reworded as "*My partner's lack of expression takes away from sharing in the feel of the moment*". Future investigations in this area might also explore stigma as one of the impacts of FM in public settings, and other contributors to FM problems in close relationships within the home. Suggested topics for future revisions of the FMQ might include fear about the progression of FM perceptions of how close family versus casual acquaintances react to FM, and feelings of stigma in public places or social gatherings. New items for PwP might include the following: "*I feel self-conscious about my*

facial appearance at gatherings with colleagues, friends or wider family"; "I am troubled by the way my partner reacts to the changes in my face"; "*I am concerned what losing expression on my face will mean for the future*". New items for SO might include the following: *"When out in public, I am bothered by the loss of expression on my partner's face";* "*I am concerned about my partner losing expression in their face in future"; "The blank response in my partner's face is frustrating"; "I find it hurtful when my partner's face does not respond".*

Some items that reflected conceptually important themes from the interviews did not behave as expected in the preliminary analysis and were subsequently removed from the factor structure. For instance, FMQ-SO #13, "*My partner's lack of expression is a barrier between us".* As the theme of distance and disconnection figured prominently for several interviewees and in respondent's open answer responses on the FMQ, this topic is worthy of further exploration.

Ambiguity in Wording of Item Statements. During expert consultation, short statements were added to demarcate topics and remind respondents to answer keeping in mind their facial expressivity. However, items could be amended to ensure they explicitly refer to FM. For example, comprehensibility of item #8 on the FMQ-PWP could be improved by changing "*It troubles me that way I feel inside is different from how I look on the outside*", to "*It troubles me that way I feel inside is different from how my face looks on the outside*".

The questionnaire's psychometric properties may have been impacted by the valence of item phrasing. As the FMQ measures a negative attribute, it included negatively phrased statements and positively phrased statements, with the positive statements being reverse scored. The practice of reversing the phrasing of items can sometimes be problematic for internal consistency and factor structures of a questionnaire (Barnette et al., 2000). In addition, the absence of FM-difficulty does not mean an equivalent absence of psychological distress or relationship strain. It is possible that some items of the FMQ could be capturing general relationship state impressions, rather those likely to related to FM. Combined with the point above, future investigations could test a version of the FMQ with a

consistent item-phrasing valence. This may result in questionnaire that has greater psychometric properties and may also be more likely to lessen the burden of cognitive switching for respondents.

Fluctuations in FM Severity. General feedback provided by respondents indicated the FM severity item may not capture daily fluctuations of this symptom. For instance, some PwP reported FM varied due to factors such as fatigue and medication off-periods, stating their appearance could range from close to normal, to almost no expression. An understanding of state vs trait-like dimensions of FM might be captured by re-phrasing the FM severity item of the questionnaires to ask respondents to rate FM severity “when facial movement/expression is at its worst” and then “as facial movement/expression appears most typically”. As the remaining FMQ subscale questions include a response scale that captures frequency, these may already take account of fluctuations in FM severity.

Study 2 Appendix VI: Final Questionnaire Versions and User Instructions

FMQ-PwP (Final version)

Please carefully read the instructions. If you find anything unclear or need assistance, talk to the administering person. There is no time limit.
Do not think too long on your replies, your immediate answers are often best.

Name: **Gender:** Male / Female / Non-binary / Prefer not to say
Date:

In Parkinson's there is a loss of mobility in the muscles of the body as well as the face. This questionnaire helps estimate the level of movement and expression in the face. It also asks about common problems of having reduced facial movement. This questionnaire can identify people who may benefit from additional information and/or support.

If you make a mistake or want to change an answer, "x" it out and mark your new answer. If you have difficulty writing or find anything unclear, please speak with the person administering the checklist.

1	**Please rate the overall expression or movement of your face, on everyday basis. Tick the answer which best describes your facial appearance, most of the time.**	Tick **one**:
	Facial movement and/or expressions are almost always absent. For example: Your face seems fixed (looks the same much of the time). A smile or other emotional expression is rare. You may look blank or as if staring often.	
	Facial movement and/or expression are noticeably less intense or less frequent than before Parkinson's developed. For example: smiling may be infrequent, look more like a grimace, be smaller, slower or delayed. You may look blank or as if staring at times.	
	Facial movement and/or expressions appear somewhat less intense or less frequent than before Parkinson's developed. For example: expressions may appear 'watered down'. You may occasionally look blank or as if staring occasionally.	
	Facial movement and/or expressions are present and unchanged. For example: your face appears as mobile and animated as prior to the development of Parkinson's symptoms.	

		Circle **one**:		
2	**Had you heard about the symptom of facial masking (the loss of facial expression or movement) before?**	I have heard of it and feel informed	I have heard of it, but do not know much about it	I have not heard of it before today

Looking back over the *past month,* help us to understand how reduced facial expression might have affected you and your relationships.

Read each question and tick the answer which best describes your current situation.

Answer once for each item. If you make a mistake or want to change an answer, "x" it out and tick your new answer. If you have difficulty writing or find anything unclear, please speak with the administering person. These questions were developed for use with people who have Parkinson's disease and their spouse or partner but can also be used with adult whānau and friends. If this is you, please read the word 'partner' with the appropriate relationship descriptor.

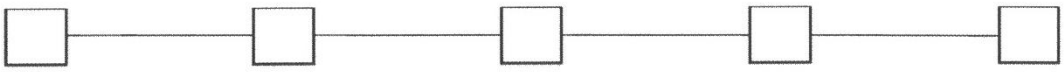
As you answer, keep in mind your level of facial expression.						*Admin only (Do not mark below)* ID RD OCI
		Rarely/ Never	**Sometimes**	**Often**	**Almost all the time**	
3	**When I am happy or pleased, people close to me can easily tell what I am feeling**					R
4	**People comment I appear to be in a bad mood much of the time.**					
5	**When I try to make an expression like a smile, it takes a lot of effort, feels false, or "put on".**					
6	**It troubles me that my face does not look like me.**					
7	**It troubles me that way I feel inside is different from how I look on the outside.**					
8	**My partner misreads my emotions, attitude, or communication**					
9	**My partner is supportive and understanding when I need to talk or confide in them.**					R
10	**I withdraw from people close to me because of my facial appearance.**					
11	**People have been unkind to me because of my facial appearance**					
12	**I avoid interactions with others because of what they might think of my face.**					

As you answer, keep in mind your level of facial expression.						Admin only (Do not mark below) ID RD OCI
		Rarely/ Never	Sometimes	Often	Almost all the time	
13	I feel isolated from people important to me because they cannot see emotion on my face.					☐
14	I take extra medication to make my face more expressive in social situations					☐
15	My partner is supportive and understanding when I need to talk or confide in them.					R ☐
16	My partner listens to me.					R ☐
17	My partner avoids or ignores me because my face does not respond.					☐
18	Interactions with my partner can feel uncomfortable or stilted.					☐
19	My lack of expression creates a barrier with my partner.					☐
20	I feel loved and wanted by my partner.					R ☐
21	My romantic relationship with my partner is strong and rewarding					R ☐
Now, a few questions about your mood and wellbeing						TOTAL ID: RD: OCI: ☐ ☐ ☐
		Rarely/ Never	Sometimes	Often	Almost all the time	(Admin only) MH
22	I feel uncertain, worried, or concerned.					☐
23	I feel angry, irritated, or frustrated.					☐
24	In general, I feel happy or in good spirits.					R ☐

25. Please briefly describe any strategies you use to cope *with reduced facial expression/movement*. State how helpful, or not helpful, you have found these strategies.

26. Take a moment to reflect on what living with reduced facial expression/movement is like *for you*. Please briefly describe your experience here:

27. Please rate how the loss of facial expression/movement has affected *you* and *your relationships.* Tick the box which best describes your current situation.



Not at all | A little | Moderately | Quite a lot | Considerably

28. Would you like more information about the topics raised in this questionnaire?

☐ Yes ☐ Not at the moment

FMQ-SO (Final Version)

Please carefully read the instructions. If you find anything unclear or need assistance, talk to the administering person. There is no time limit.
Do not think too long on your replies, your immediate answers are often best.

Name: **Gender:** Male / Female / Non-binary / Prefer not to say
Date:

In Parkinson's there is a loss of mobility in the muscles of the body as well as the face. This questionnaire helps estimate the level of movement and expression in the face. It also asks about common problems of having reduced facial movement. This questionnaire can identify people who may benefit from additional information and/or support.

If you make a mistake or want to change an answer, "x" it out and mark your new answer. If you have difficulty writing or find anything unclear, please speak with the person administering the checklist.

1	**Please rate the overall expression or movement of the person with Parkinson's face, on everyday basis. Tick the answer which best describes their appearance, most of the time.**	Tick **one:**
	Facial movement and/or expressions are almost always absent. For example: The face seems fixed (looks the same much of the time). A smile or other emotional expression is rare. The face may look blank or as if staring often.	
	Facial movement and/or expressions are noticeably less intense or less frequent than before Parkinson's developed. For example: smiling may be infrequent, look more like a grimace, be smaller, slower, or delayed. The face may look blank or as if staring at times.	
	Facial movement and/or expressions appear somewhat less intense or less frequent than before Parkinson's developed. For example: expressions may appear 'watered down'. The face may occasionally look blank or as if staring occasionally.	
	Facial movement and/or expressions are present and unchanged. For example: the face appears as mobile and animated as prior to the development of Parkinson's symptoms.	

		Circle **one**:		
2	**Had you heard about the symptom of facial masking (the loss of facial expression or movement) before?**	I have heard of it and feel informed	I have heard of it, but do not know much about it	I have not heard of it before today

Looking back over the *past month*, help us to understand how reduced facial expression might have affected you and your relationships.

Read each question and tick the answer which best describes your current situation.

Answer once for each item. If you make a mistake or want to change an answer, "x" it out and tick your new answer. If you have difficulty writing or find anything unclear, please speak with the administering person. These questions were developed for use with people who have Parkinson's disease and their spouse or partner but can also be used with adult whānau and friends. If this is you, please read the word 'partner' with the appropriate relationship descriptor.

	As you answer, keep in mind the level of facial expression you see					*Admin Only (Do not mark below)* FMD OCI
		Rarely/ Never	**Sometimes**	**Often**	**Almost all the time**	
3	**My partner appears to be in bad mood a lot of the time** For e.g., sad, grumpy, bored, cold, disengaged, or uninterested.					☐
4	**It is hard for me when my partner's face doesn't respond.**					☐
5	**My partner shares in my life's ups and downs.**					☐ R
6	**I can easily tell when my partner is feeling positive emotions.**					☐ R
7	**My partner is more expressive with other people, than with me.** For e.g., they try to smile at a social event but not when we are at home alone.					☐
8	**I find myself second-guessing what my partner is thinking and feeling.**					☐
9	**I think my partner's unhappiness must be my fault.**					☐
10	**I can count on my partner to be supportive and understanding.**					☐ R

As you answer, keep in mind the level of facial expression you see						*Admin Only (Do not mark below)* FMD	OCI
		Rarely/ Never	Sometimes	Often	Almost all the time		
11	I feel rejected or criticised when my partner shows little expression.					☐	
12	My partner is interested in interacting with me.					R	
13	I avoid being near my partner or interacting with my partner.						☐
14	I have a close and warm relationship with my partner.					R	
15	It is upsetting my partner's face/expression has changed so much from their former self.					☐	
16	I remind myself how my partner's face looks on the outside is probably not how they feel inside.					☐	
17	I feel disconnected from my partner because their face does not respond to me.					☐	
18	My partner's emotional signals make me concerned they might want to end our relationship.						☐
19	My partner's emotional signals make me concerned they are considering an affair or are having an affair.						☐
20	I feel a strong emotional connection with my partner.					R	
21	My partner's staring or lack of expression gets in the way of our sexual relationship.						☐
22	I think my partner might be better off without me.					☐	
23	I worry my partner cares less for me-than before.					☐	
24	My romantic relationship with my partner is strong and rewarding.						R

		Rarely/ Never	Sometimes	Often	Almost all the time	*Admin Only FMD OCI*
25	I wish I were closer to my partner.					☐
26	My partner makes me feel loved and wanted.					R ☐
	Now, a few questions about your mood and wellbeing					TOTAL FMD: ☐ ☐
		Rarely/ Never	**Sometimes**	**Often**	**Almost all the time**	*Admin Only MH*
27	I feel uncertain, worried, or concerned.					☐
28	I feel angry, irritated, or frustrated.					☐
29	In general, I feel happy or in good spirits.					R ☐

30. Please briefly describe any strategies you use to cope *with reduced facial expression/movement*. State how helpful, or not helpful, you have found these strategies:

31. Take a moment to reflect on what living with reduced facial expression/movement is like *for you*. Please briefly describe your experience here

32. Please rate how the loss of facial expression/movement has affected *you and your relationships.* Tick the box which best describes your current situation.

☐ Not at all ☐ A little ☐ Moderately ☐ Quite a lot ☐ Considerably

33. Would you like more information about the topics raised in this questionnaire?

☐ Yes ☐ Not at the moment

FMQ USER INSTRUCTIONS

The FMQ can be administered in pen and paper form or verbally, ideally as part of an assessment interview once rapport has been established. The FMQ-SO was primarily developed for use with the spouse or partner of people who have PD, but it may also be used with adult family members or close friends. A significant other may also complete the questionnaire alone, with consideration given to how the person who has PD may be included in the assessment and/or discussion of findings. Assistance should be provided to patients who have difficulty writing their responses or would find writing unduly taxing. If administered verbally, the questionnaire should be undertaken in a private room or office because of the sensitive nature of some of the items.

Since people with PD and their families may not realise FM is a symptom and/or may be reluctant to raise the topic with their healthcare providers, it is recommended that items 1 and 2 of the questionnaires are administered even when FM is not seen on clinical exam (these items may be abbreviated to be conducted verbally). If FM is perceived to be present by one or both respondents, it is recommended to proceed with the remainder of questionnaire to evaluate the impact of living with FM, at the administrator's discretion.

SCORING SUMMARY

The FMQ provides an **FM severity rating, FM-related difficulty subscales** (two subscales for PwP and a single scale for SO), and a **total score.** Additional items provide clinically or conceptually important information to the administrator, including a mental health screen, open answer items assessing subjective experience and self-management of FM, perceived global FM Difficulty rating, and FM education preference.

- ***FM Severity & Awareness Rating***

  Item 1 requires the respondent to select the description which best describes their/their partners level of facial expressivity (or facial masking). No calculation is required. The topmost response option corresponds to Severe

FM, with the next lowest option indicating Moderate FM, the following option Low FM, and the bottom description indicates normal expression.

Item 2 requires the respondent selects the description which best describes their FM knowledge. No calculation is required.

- ***FM-related Difficulty Subscales***

  Respondents indicate the frequency each item is experienced. Items are scored as following: Rarely/Never = 0, Sometimes = 1, Often = 2, Almost all the time = 3. Simply write the numeric value corresponding to respondent answers for each item, in the box on the righthand side of the questionnaire. When each item score has been completed, add up the numeric scores for each of the vertical columns to calculate the raw scores.

  FMQ-PwP Individual Difficulty (ID) subscale

  Summed Items: 5, 6, 7, 10, 12

  FMQ-PwP Relationship Difficulty (RD) subscale

  Summed Items: 4, 15, 16, 17, 18, 20.

  FMQ-SO FM Difficulty (FMD) scale

  Summed Items: 3, 4, 5, 6, 8, 9, 10, 11, 12, 14, 15, 16, 17, 20, 22, 23, 25, 26.

  Please note scoring is reversed for FMQ-PWP items 15, 16, 20 and FMQ-SO items 5, 6, 10, 12, 14, 20, 26.

- ***Other Clinical Indicators***

  This is a set of items that represent clinically or conceptually important topics, but due to their psychometric properties are separate to the main subscale items. Other Clinical Indicator (OCI) items are scored on the same response scale as the FM Difficulty subscales (above). *Please note scoring is reversed for FMQ-PWP items 3,9,21 and FMQ-SO item 24.*

- ***FMQ Total Score***

For the *FMQ-PwP* a total score is calculated by adding Individual Difficulty (ID) subscale, the Relationship Difficulty (RD) subscale, and the set of items forming Other Clinical Indictors (OCI). For the *FMQ-SO*, a total score is calculated from adding the FM Difficulty scale (FMD) to the set of OCI items.

Other items of note:

- ***Mental Health Screen***

  There are three separate items which provide a general screening of psychological distress (FMQ-PwP items 22-24, and FMQ-SO items 27-29). These are also scored on the same item response scale as FM Difficulty subscales (above). Any individual item score of 2 or higher signals the need for further investigation or psychological/specialist assessment. *Please note scoring is reversed for FMQ-PWP item 24, and FMQ-SO item 29.*

- ***Open Answer & Single Index Items***

  There are two open answer items which provide information on the experience of having FM and how well any socioemotional difficulties arising from having FM are self-managed. Responses to these items offer subjective information to gauge how impactful FM is as a symptom of PD and help to identify intervention targets. There are two further single items, one where global FM difficulty is rated on a 5-point scale, and another item indicating health education preference in a yes/no format. These single index items require no calculation and are included to assist with score interpretation and patient education recommendations.

INTERPRETATION OF SCORES

The higher the score on the FMQ subscales and total score, the more subjective wellbeing is compromised. There are no cut-off scores to indicate clinical significance.

- **FMQ-PwP Total Score**

  Theoretical range: 0 - 60, Observed range during initial validation: 0 - 27
- **FMQ-PwP Individual Difficulty (ID) subscale**

  Theoretical range: 0- 18, Observed range during initial validation: 0-15
- **FMQ-PwP Relationship Difficulty (RD) subscale**

  Theoretical range: 0- 18, Observed range during initial validation: 0-12
- **FMQ-SO Total Score**

  Theoretical range: 0-72, Observed range during initial validation: 0-49
- **FMQ-SO FM Difficulty (FMD) scale**

  Theoretical range: 0-54, Observed range during initial validation: 0-40

Interpretation of the results can also be aided by subjective comparison of scores across respondent versions, and within the questionnaire. Administrators may consider the following elements when interpreting the FMQ:

- Discrepant FM severity ratings between PwP and SO may indicate poor awareness of FM as symptom of PD, or poor insight of the degree of facial expressive impairment.
- A large disparity in FM difficulty scores between PwP and SO may occur (one respondent scoring exceptionally low and the other high on FM difficulty subscales, or as subjectively judged for the open answer and single index items). This indicates a difference in the consequences of FM between respondents, or in how they may be coping with FM. It may be helpful to discuss if one or both individuals are unaware the impact FM has on the other.

- If a respondent indicates low FM severity or normal expression, but has elevated scores on the FM difficulty subscales, this may also reflect low awareness or insight of FM. Objective assessment of FM can be confirmed by clinician-based observation or use of other clinical staging tools. On the other hand, if an administrator *does not* observe signs of FM (such as reduced expressivity, reduced blink rate, or a lack of active mouth closure) this may reflect problems associated with of another aspect of PD (such as speech difficulty). In the latter circumstance, it is recommended administrators enquire why a respondent positively to the items contributing to the elevated FM difficulty scale score.

RECOMMENDATIONS FOR PATIENT EDUCATION/SUPPORT

Engaging in a dialogue to ascertain awareness of FM and discuss its potential consequences is generally encouraged regardless of the apparent clinical staging of FM pathology seen on exam. The higher the FMQ scores, the more strongly education and support are recommended. Practitioners should use their clinical judgement to decide the depth and content of information appropriate to each respondent. Decisions to provide information can also be guided by responses to the symptom knowledge and education preference items.

Responses to the open answer items will assist administrators in understanding of what living with FM has been like for that individual and help identify intervention targets. For instance, a respondent who reports minimal strategies to help them cope with FM may benefit from referral to physiotherapy or speech programmes – which may mitigate the reduction of facial mobility in the short term. Respondents who report relationship strain related to FM may benefit from tips to strengthen connection and support communication without facial expression. For example, verbalising what is felt for the person with PD, and for family members, relying on what is said and done rather than guessing or looking at the

face. A summary of compensatory behaviours used by people with FM and their partners can be found in Wootton, Starkey, and Barber (2018), but note that further investigation is required of what is most helpful and for whom.

6. Study 3
Face Value: Perception of Facial Masking and the Link to Psychosocial Outcomes in Parkinson's Disease

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Face Value: Perceptions of Facial Masking and the Link to Psychosocial Outcomes in Parkinson's Disease

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Word count: approx. 5798 (abstract: 249)

Author Note

The findings of this paper form part of a wider doctoral research project, thus participant characteristics and method sections of the present report have similarities to work previously published from that project.

Abstract

Background: Little is known about how people with Parkinson's disease or their family members perceive facial masking, or the effects of this symptom on psychosocial outcomes. This study explored the psychosocial correlates of facial masking for people who have Parkinson's and their family members. A secondary aim was to compare how different observers perceived facial masking.

Methods: Self-report questionnaires measuring psychological health, social functioning, and quality of life were completed by 80 people who had Parkinson's and 58 of their significant others. Self and significant other ratings of facial masking severity were made on the Facial Masking Questionnaire (FMQ). Clinical ratings of facial masking severity were made by two healthcare professionals using the MDS-UPDRS, based on videos of the participants, and the researcher (AW) coded facial expressive behaviours from the same video.

Results: Greater perceptions of facial masking severity/lower retained expressivity were significantly associated with greater masking-related difficulty, which was in turn strongly associated to poorer psychosocial outcomes. Clinical ratings of facial masking were not significantly related to people with Parkinson's or significant other reports of facial masking severity. Few significant correlations were found between healthcare professional ratings of masking-related difficulty, or with psychosocial outcomes.

Conclusion: Facial masking can have detrimental effects on psychosocial outcomes, but individuals with Parkinson's and their family members may be affected differently. Relying solely on clinical ratings of facial masking may not accurately appraise the possible effects of this symptom in the everyday lives of people with Parkinson's and their families.

Keywords: Parkinson's disease, facial masking, hypomimia, psychosocial outcomes.

Facial masking (FM) is a characteristic and common symptom of PD [1, 2]. FM involves a decrease in facial emotional expressivity and an unusual stillness in the face, arising from disease-related impaired mobility and increased rigidity of the facial musculature. It has been long noted that FM leads people with PD to be perceived as losing feeling, interest, or intellect [3]. Research examining social perceptions of people with FM has shown perceivers form negatively biased judgements of mood, character, and mental state [4-8]. People are also less accurate at recognising the emotions of people with FM [9]. Even healthcare practitioners with knowledge of PD may find it difficult to override the negative impression that FM appears to produce. When shown silent videotapes of people with heart disease and people with PD, healthcare professionals made more negative judgements of intellect, affect and personality traits in the individuals with PD, despite there being no other measured difference between the groups [10]. As the degree of expressive loss increases, individuals with FM tend to be evaluated less favourably by healthcare professionals, especially in interpersonal and affective domains [4, 6]. Unsurprisingly, less experienced (student) practitioners were found to be more susceptible to forming inaccurate judgements of individuals with FM, compared to practitioners with greater experience in PD, although neither seem to be immune to the influence of FM [4]. Interestingly, the amount of expressivity displayed in PD may be, in part, be a function of the type of relationship or the social setting. Individuals with PD were observed to display greater expressivity when interacting with an attentive but unfamiliar person, such as a researcher, compared to when conversing with their spouse in the same environment [11]. Thus, the effects of expressive loss may be different when the perceiver is more familiar with the individual with PD. While several studies have explored how FM affects social perception in casual or healthcare interactions, little is known of how people with PD themselves and their partners perceive FM, or how their perceptions of this symptom might correspond to psychosocial outcomes in PD. These topics are the focus of the present study.

PD is now recognised to affect much more than physical mobility; it deeply affects an individual's daily life [12]. Motor symptoms may cause difficulty with walking and carrying out tasks of everyday living, while problems in communication, cognition, behaviour, emotion, sleep, fatigue, and pain may also compromise quality of life (QOL) [13]. One factor thought to contribute to QOL in PD is social functioning [14-16]. Difficulties in speech and communicating [17], feelings of social isolation [18], stigma [19], and changes to the ability to read social cues and emotion [20] are thought to have strong implications for social functioning in PD.

PD does not just happen to the individual; it also affects those who share in their life. Family and couple relationships are influenced from the earlier stages of the condition, as changes to the individual's functional ability reshape roles and responsibilities in family systems [20]. Caregiving for a person with PD is also associated with decreases in psychological and physical health [21, 22]. Given that the visible displays of affection, care and understanding may become compromised in FM, deepening understanding of how FM may influence relationships with family and carers is important, to identify opportunities that promote satisfying and supportive relationships and to help people better cope with the challenges and changes that occur in PD.

Few clinical evaluation tools include FM. The Movement Disorder Society's Unified Parkinson's Disease Rating Scale (MDS-UPDRS) provides clinical staging of PD symptoms, including FM [23], but it does not include questions to assess FM-related psychosocial dysfunction. The Parkinson's Disease Questionnaire (PDQ-39) [24], a commonly used measure of QOL in PD, includes three questions about social functioning but none refer to FM [25]. Computer and video-based techniques have also been investigated as methods of identifying FM [26, 27].

Only a few studies have investigated how FM relates to psychosocial outcomes in PD. Qualitative research has shown FM is perceived as embarrassing [28] and may cause distress between couples when it is inaccurately viewed a diminishment of emotional

attachment [29]. Similarly, our earlier work suggested partners of people with FM have trouble attributing the symptom of FM to PD, seeing it as persistent bad mood or impassiveness, resulting in a loss of feelings of security and confidence in their relationship [30]. Two quantitative studies have investigated the effects of FM in PD. Gunnery and colleagues [31] found greater levels of FM corresponded with decreased relationship enjoyment for partners, but not people with PD. For the participants with PD, having FM predicted increased feelings of social rejection, although this association was reduced when controlling for depression [31]. In Ma and colleagues [32], greater feelings of stigma and female gender were found to mediate poorer quality of life in people who experience FM. It is not clear if the findings on stigma extend from general social functioning to primary social relationships. Psychosocial functioning is important to address clinically because PD is an incurable condition characterised by increasing disability, with most individuals experiencing a considerable reduction in functioning across all areas of quality of life [33, 34]. With day-to-day support often relying on familial caregivers, it is important to document the psychosocial consequences of the condition for individuals with PD and their family members.

Purpose of the Present Study

This paper focuses on the effects of FM on psychosocial outcomes in people with PD and their significant others (spouse/partner or other close family member). The main aim was to assess the association between FM and psychosocial outcomes for both people who have PD and their family members. The secondary aim was to compare how different observers perceived FM.

Method

Participants

This research included 138 community-residing participants from the North Island of Aotearoa New Zealand, comprising people who have PD (PwP, *n* = 80) and their significant others (SO*, n* = 58). Demographic and clinical characteristics for the participants are displayed in Table 1. Advertisements seeking participants were placed in PD support group newsletters and emailed to support group members. Data collection took place from March to November 2019. After potential participants made contact, they were telephoned and screened for eligibility. Verbal/written information about the study was provided to potential participants, before being asked if they consented to taking part. Testing usually took place at the participant home or workplace.

Individuals were required to have been diagnosed with idiopathic PD by a healthcare professional, usually a neurologist. Respondents who might have required extra support, such as those formally diagnosed with a memory impairment or who had difficulty conversing in English, were excluded from taking part. As we did not want to exclude people with PD who had no available significant other, individuals with PD could also take part by themselves. Selection was not made based on FM status, so that participants had a range of FM severity from none to severe.

Design

This cross-sectional multiple centre project forms part of a wider programme of research and a portion of the participant characteristics and method sections described here are similar to that published previously [35].

Assessment and Procedure

Four separate groups provided ratings of FM severity using various methods of measurement (PwP, SO, healthcare professional, and researcher). Self/SO-reported expressive disposition of the PwP was also measured. Psychosocial outcomes assessed in PwP included psychological health (symptoms of anxiety and depression), social functioning

(marital satisfaction and affectionate/positive aspects of social support), and health-related quality of life. Psychosocial outcome measures completed by SO included psychological health (general psychological health and caregiver strain), and social functioning (general social relationships quality, marital satisfaction, and affectionate/positive aspects of social support). The assessment session included the PwP/SO completion of self-report questionnaires and a video recording of a discussion approximating natural interaction between the PwP and the researcher (AW).

Ethical approval was received from the University of Waikato School of Psychology Human Research Ethics Committee (#2018:46). Respondents received verbal and written information about the study and gave written informed consent before participating. At the completion of questionnaires, consenting participants received a $20 supermarket voucher. The videoed discussion was a separate and optional consent, so individuals who were sensitive about having their facial movement recorded/analysed could opt out without impacting their overall participation. Five participants opted out of the video, and an additional two videos were unable to be analysed, as Parkinson's related symptoms made viewing the face difficult. As the New Zealand Parkinson's community is relatively small, participants were informed their video might be reviewed by an expert known to them and were given the option to nominate their own preferred healthcare professional to view the recording. One participant chose this option, but that video was one of the two unable to analysed.

Self-report Measures

Social functioning was measured using Medical Outcomes Study-Social Support Survey (MOS-SSS) [40]. For the sake of brevity and relevance, we selected the six items comprising affectionate support and positive interaction subscales [41]. Affectionate support items measure perceived expressions of love and affection, where positive social interaction items measure perceived availability of other persons to share in enjoyment or in fun activities. Items are answered on a five-point scale from 1 (None of the time) to 5 (All of the

time), with lower scores indicating a lower level of perceived social support. These two subscales have been previously modified to assess spousal relationship enjoyment in PD [31]. We used the original items, asking study participants to rate their most important (closest) social relationships, to avoid excluding those who took part without a romantic partner. Participants in marital/stable relationships also rated relationship satisfaction using the Kansas Marital Satisfaction Scale (KMSS) [42]. This brief three-item scale measures how satisfied an individual is in their relationship. Items are rated on a 7-point scale, ranging from extremely dissatisfied (1) to extremely satisfied (7). Higher scores indicate greater marital satisfaction/quality, with a marital distress/non-distress cut-off score of 17 [43].

To measure psychological health, PwP completed the Hospital Anxiety and Depression Scale (HADS), a 14-item screening tool which measures symptoms of anxiety and depression in hospital and community settings [44]. Each item is rated on a four-point scale. The HADS is shown to be a valid and responsive scale for PD populations [45], with a score of 11 or higher indicating clinical distress in PD [46].

Disease-specific quality life measures exist for PD but had a prohibitive cost for this project. Instead, PwP completed the Health-Related Quality of Life Instrument: Fifteen Dimensions (15D) [47]. This 15 item self-report questionnaire utilises population-based preference weights to produce a single index score on a 0 to 1 scale, where 0 = being dead and 1 = no problems on any dimension. The item response format is a 1-5 scale, with the respondent choosing the best descriptor of their present state of health. The 15D has been shown to be a valid instrument to assess Health-Related Quality of Life (HRQOL) in PD populations [48].

SO psychological health was assessed using the World Health Organisation Quality of Life Assessment -Short Version (WHOQOL BREF) [49]. This 26-item measure four broad domains, each providing a score: physical health, psychological health, social relationships, and environment. Items are rated on a five-point Likert-type scale, with scores calculated on a 100-point scale (where higher scores indicate better QOL). This questionnaire is frequently

used in patients and caregivers in various chronic illness and has been previously used in PD caregiver populations [50]. Psychological health in SO was also assessed with the Caregiver Strain Index (CSI) [51]. The CSI a 13-item self-report questionnaire that measures burden strain related to the caregiving role in long-term family caregivers. Strain is assessed across five domains: Financial, Physical, Psychological, Social, and Personal. The response format is yes/no, with higher scores indicating greater caregiver burden. Although there is a newer version which uses a 0-2 response scale, the original was used here to aid comparison with previous usage in Parkinson's caregivers [52-54]. The range of mean CSI scores across those PD studies is reported as 3.0 – 8.7 (SD 2.7 – 3.5) [55].

FM variables were assessed using the Facial Masking Questionnaire (FMQ), an instrument designed to assess the amount of psychosocial difficulty associated with facial masking in people who have PD and their significant others [35]. The FMQ-PwP (person with Parkinson's version) is a self-report scale (*n* = 28 items) of FM-related difficulty that includes individual and relationship subscales. The FMQ-SO (significant other version) is a unidimensional self-report measure (*n* = 30 items). Items are rated by selecting how often each statement occurs on a four-point response scale where 0 = rarely/never and 3 = almost all the time. Several additional items of clinical relevance are included: perceived FM severity, knowledge of FM, indicators of psychological distress, open answer items to voice personal experiences of FM, a global FM difficulty rating item, and preference for further information about FM.

Individual expressive disposition was assessed using The Berkeley Expressivity Questionnaire (BEQ), a 16 item self-report questionnaire measuring perceived emotional expressive disposition across three facets: negative expressivity, positive expressivity, and impulse strength [41]. BEQ items are rated on a 7-point scale, from strongly disagree to strongly agree. Higher scores show greater perceived expressiveness. We administered the BEQ to PwP participants and created a modified peer-report version for SO participants to complete.

Videoed Discussion

An informal discussion between the first author and the participant was recorded. Usually, this was conducted after completing the self-report measures when participants had established a rapport with the researcher. During the videoed discussion, the researcher asked five questions which were chosen to increase the likelihood of range of emotional expressions being elicited and to provide balance to the emotional tone of the interaction. Question topics included: Q1) typical daily routine, Q2) enjoyed activities, Q3) a frustrating or challenging event in the past few weeks, Q4) an enjoyable event in the past few weeks, and Q5) a future event the interviewee is looking forward to. The questions were followed by informal prompts to increase elaboration, where necessary. Four of the five questions topics (Q1, Q2, Q3, & Q4) were adapted from existing interview procedure developed for the Interpersonal Communication Rating Protocol – Parkinson's disease version [56].

Raw video data was transformed by removing audio (to minimise the influence of verbal communication) and then edited into two sets of silent clips. The first set of silent clips was rated by the first author and a research assistant to provide assessment of facial expressive movement and key clinical features of FM. Response start and end times for the five interview questions were identified and standardised clips were extracted by taking the first 20-45s of each response. Extraction length was calculated separately for each question, by retaining the maximum duration while keeping the number of participants with insufficient footage to <15% for each question. Extracted clips for all five questions were then coded (around 170s total duration per participant). Raters used the Facial Expression Coding System (FACES) to measure the frequency and duration of facial expressive behaviours [57, 58]. In addition to expressions of emotion, codes were added for 'neutral' (at rest with no observable expression and no observable features of FM) and 'masked' (one or more of: reduced blink frequency; unintentional lip/jaw separation or lips closed with jaw visibly dropped and unintentional downturned lip corners; eyebrows which may be raised or furrowed but are unusually 'stuck'; a generally impassive rather than neutral appearance).

Neutral and masked codes were combined for a researcher-based rating of facial masking. The first author and a research assistant trained as raters on the modified version of the FACES, using the unextracted video data. Raters trained on 20% of the sample (*n* = 15) until an acceptable level of inter-rater agreement was reached on all observation pairs (*kappa* >.7). Remaining coding using the modified FACES was then completed by the first author. Training video data was selected to represent a range of expressive dispositions and roughly equal genders of participants. Following procedure for observational video data [59], Kappa statistic was used to assess inter-rater reliability of the codes for each observation pair. Average kappa, tallying instances of all main codes by duration, was .82 (*range* = .71 to .93) and by tallying the frequency of all main codes was .77 (*range* = .76 to .81). The adapted version of the FACES and rater training protocol is described in the supplementary data.

A second set of shortened clips was created for experts to rate, so that video footage could be viewed within approximately one hour. Standardised extracts (20s in total per participant) were comprised of interviewee responses to questions 2 and 3, which were selected to maximise the likely emotional range displayed by video participants. Clips for each participant were separated by a black screen listing the clip number and instructions for the viewer to make a rating. Experts, two registered nurses experienced in providing PD support, viewed the recordings of consenting participants in person and rated the level of FM using item 3.2 of the Movement Disorder Society Unified Parkinson's disease Rating Scale (MDS-UPDRS) [60]. This clinical staging measure assesses FM severity by reduced eye-blink frequency, loss of facial expression/spontaneous smiling, and frequency of parting of lips when the mouth is at rest. The response format is a five-point scale from 0 = none, to 4= severe FM.

Data Analysis

Qualtrics Survey Software was used for data entry of paper-based self-report measures. Statistics were calculated using IBM SPSS (version 26). Encoding of video extracts was automated using PowerShell and Adobe Media Suite. Noldus Observer XT

software aided coding of facial expressive behaviours in the video data. As this is a clinical sample where much of the data had non-normal distributions, the analyses primarily use nonparametric statistics.

Spearman's rank order correlations were used examined the associations between different ratings of FM severity. Krippendorff's Alpha (K alpha) was conducted to measure the magnitude of agreement between PwP-SO pairs and between healthcare professional (Parkinson's nurse) raters [36, 37]. The versatility of K alpha was important for the present study, which involved unequal group sizes and some missing data [38]. K alpha ≥ .67 was taken as the lowest acceptable limit of agreement [36]. Upper and lower limits reported are 95% confidence intervals. Spearman's rank order correlations were calculated to explore the associations between FM variables and psychosocial outcomes. The magnitude of correlations was interpreted using Cohen's criteria [39]. Comparison of differences in observed mean psychosocial outcome scores for the present sample with other population groups (PD and nonclinical) was also carried out.

Table 1

Sociodemographic and clinical characteristics of study participants

PwP Participants (*n* = 80)	
Age in years mean (SD) range	69.64 (9.36) 44.00-86.00
Male n (%)	57.00 (71.25%)
Early Onset < 50 years n (%)	6.00 (7.50%)
Time since diagnosis mean (SD) range	6.40 (4.99) 0.50-22.00
Facial Masking duration mean (SD) range	2.63 (4.33) 0.10-23.00
Dopamine Replacement therapy n (%)	74.00 (92.50%)
No medication n (%)	4.00 (5.00%)
Deep Brain Stimulation n (%)	3.00 (3.75%)
New Zealand or Other European n (%)	73.00 (91.25%)
Māori & Pasifika n (%)	5.00 (6.25%)
Other ethnic background n (%)	3.00 (3.75%)
Retired or unable to work n (%)	65.00 (81.25%)
Married/Living together n (%)	68.00 (85.00%)
Took apart alone n (%)	22.00 (27.50%)
Lives in private home n (%)	73.00 (91.25%)
SO Participants (*n* = 58)	
Female n (%)	45.00 (77.59%)
Relationship length mean (SD) range	41.97 (12.52) 11.00-62.00
New Zealand or Other European n (%)	55.00 (94.83%)
Other ethnic background n (%)	2.00 (3.45%)
Māori n (%)	1.00 (1.18%)
Retired n (%)	40.00 (68.97%)
Has current illness n (%)	9.00 (15.52%)
Tertiary Education n (%)	31.00 (53.45%)
Took part with spouse or partner n (%)	55.00 (94.83%)
Took part with parent n (%)	3.00 (5.17%)

Note. Dopamine Replacement therapy, Sinemet or Madopar use

Results

Ratings of Facial Masking

There was a statistically significant, moderate positive correlation between PwP and SO ratings of FM severity. The more FM perceived by PwP, the more FM SO also perceived (r = .430, p = 0.01). While the correlation between PwP and SO ratings of FM severity was statistically significant, the agreement between PwP-SO pairs did not reach an acceptable level (α = .41, *lower limit* = .21, *upper limit* = .58, where $\alpha \geq .67$ indicates an acceptable level for agreement). A significant, strong positive correlation was found between the two healthcare professionals' ratings of FM (r = .71, p < .01). Their ratings also showed an acceptable level of agreement (α = .70, *lower limit* = .58 *upper limit* = .81). Subsequent analyses in this paper use the mean healthcare professional rating.

Correlations between healthcare professional ratings of FM severity and PwP reports of FM severity were not statistically significant (r = .17, p = 0.15), and SO reports of FM severity (r = .14, p = .30). Because different methods of measurement were used across the four rater groups, analysis of agreement was not suitable. The presence of FM yes/no responses across the four rater groups was compared, based on the subset of PwP participants who consented to take part in the video (n = 73 PwP video participants). The healthcare professionals separately rated 2.70 % (n = 2) and 8.2% (n = 6) of the video sample as having normal expression, whereas 33.30% (n = 25) of the PwP group rated themselves as having normal expression, and 14.30% (n = 8) of their SO's reported normal expression.

Table 2 reports the correlations of researcher-rated expressive facial behaviour with PwP/SO report and with healthcare professional assessment of FM severity. Correlations between PwP-reported FM severity and most aspects of researcher-rated facial behaviour were not statistically significant, except for high intensity expressions (table 2). SO ratings of FM severity had significant, small positive correlations with the "Masked" (clinical features of FM, additional to reduced expression) and "FM Severity" (combined neutral and masked)

codes (table 2).

Health professional evaluations of FM were significantly related to most researcher-rated facial behaviours, but the highest of these correlations was of only of moderate magnitude (maximum *r* = .53, “masked” code).

Table 2

Correlations of researcher-rated expressive facial behaviour with PwP/SO report and with healthcare professional assessment of FM severity.

Facial Behaviour Codes	**FM Severity by Rater**		
	Healthcare Professional	**PwP-Report**	**SO-Report**
EXPRESSION (observable emotion or expressive movement)	**-.29***	-.15	-.24
NEUTRAL (at rest with no observable expression)	**-.37****	-.21	-.17
MASKED (other clinical signs of FM)	**.53****	.18	**.30***
FM SEVERITY (Masked & Neutral combined)	**.28***	.16	**.28***
Modifier codes for type of Expression			
Positive	-.22	-.09	-.23
Negative	.19	.04	.15
High-Very High Intensity	-.14	**-.26***	-.13
Low-Medium Intensity	.11	.21	.06

Note. Spearman rank correlation coefficients. Significant correlations in bold. Facial behaviours coded by researcher from video using the modified FACES. Codes represent the proportion of observation (duration). Other clinical signs of FM in addition to reduced expression, reduced blink rate, mouth dropping open, unusually fixed eyebrows. Healthcare Professional ratings, MDS-UPDRS; PwP/SO-report, FMQ-PwP/FMQ-SO.

*Correlation is significant at the 0.05 level (2-tailed)

** Correlation is significant at the 0.01 level (2-tailed)

General Psychosocial Outcomes

People with Parkinson's

Scores on the psychosocial outcomes and comparison with the general older population and other clinical samples, are provided in table 3. PwP in our study had poorer HRQOL when compared with other PD groups and similarly aged (non-clinical) older adults, with a significant difference in mean scores between the present sample and non-clinical older adults (see table 3).

Good psychological health was generally reported by PwP. Overall, low levels of symptoms of depression and anxiety were reported (mean HADS depression score = 4.50, mean HADS anxiety score = 5.65, where score of ≥11 indicates clinical levels of distress in PD populations). Three respondents (~4%) had scores corresponding to a clinical level of depression symptoms, and 12 (15%) had scores corresponding to anxiety symptoms at a clinical level. Interestingly, psychological distress of the present sample was significantly lower than that reported in other studies of PD and in general samples of similarly aged New Zealanders (see table 3).

PwP perceived a good overall availability of positive and affectionate support from the important people in their lives (MOS-SSS positive interaction mean score = 70.25, affectionate support mean score = 78.0, where maximum score is 100 and higher scores indicate greater support). PwP in the present study had a statistically significant difference in both perceived affectionate support and positive social support, when compared with general older populations. Perceived availability of positive social interactions, but not affectionate support, was significantly lower in the present sample relative to other clinical populations.

The marital satisfaction of people with PD indicated they were not, on average, experiencing relationship distress (mean KMSS score = 19.36, where higher scores indicate greater marital satisfaction/quality with scores < 17 indicating marital distress). The present sample reported similar levels of marital satisfaction to other clinical and non-clinical populations. In summary, the present sample of PwP reported similar quality of life,

relationship satisfaction, and availability of affectionate support when compared to other clinical samples. They perceived a lower availability of positive social interactions but had better psychological health than other clinical samples.

Significant Others

SO psychosocial outcomes, comparisons with the general older population, and comparisons with other clinical samples, are provided in table 4. SO in our sample had lower QOL domain scores for psychological health and physical health, relative to their social relationships and environment scores (WHOQOL BREF domain scores, see table 4). SO physical health and psychological health (domain scores) were similar to other carer populations, but significantly worse than in general older adult samples. SO in the present sample also experienced similar levels of caregiver strain to other PD carer samples.

SO perceived a poor availability of affectionate support and positive social interaction, compared to caregivers in other clinical groups and community samples of older adults (see table 4). They were also significantly more dissatisfied with their relationships compared to general older adult populations but experienced a similar level of marital satisfaction to carers in other clinical populations (table 4). Overall, SO in the present sample reported similar levels of psychological health, caregiver strain and marital satisfaction to carers in other samples. They perceived a lower availability of affectionate support and positive social interactions than carers in other conditions.

Table 3

Descriptive statistics comparing PwP scores on psychosocial outcomes, with scores of other PD populations and nonclinical populations.

	Present Study		Other PD populations [a]						Non-Clinical Populations					
Psychosocial Outcomes	***n***	***Mean (SD) range***	***Study***	***Sample***	***Mean (SD)***	***Mean difference***	***t***	***p***	***Study***	***Sample***	***Mean (SD)***	***Mean difference***	***t***	***p***
Health Related Quality of Life	79	.73 (0.92) .47 to .93	*Haapaniemi et al (2004)*[48]	Finnish PD patients from routine care (n= 256, mean age in years= 67)	.77 (0.13)	0.04	0.68	.499	*Haapaniemi et al., (2004)*[48]	Finnish age matched controls (*n* = 256)	.86 (0.12)	0.13	2.21	**.028***
Anxiety	79	5.65 (3.96) 0 to 17	*Martinez Martin et al., (2005)*[21]	Spanish PD patients from routine care (*n* =72 patients, mean age in years= 67)	7.54 (3.90)	1.89	2.95	**.004***	*Roberts, Fletcher, & Merrick (2014)*[61]	Older New Zealand adults from a community sample (*n* = 203, mean age in years= 68)	11.9 (3.6)	6.25	12.73	**.001****
Depression	79	4.51 (2.71) 0 to 12			7.86 (0.52)	3.35	10.32	**.001****			9.9 (2.5)	5.39	15.88	**.001****
Affectionate Support	79	78.0 (1.25) 0 to 100 [b]	*Raggi et al., (2009)* [62]	Italian adults treated for autoimmune neuromuscular disease (*n* =74, mean age in years= 48)	75.2 (22.2)	-2.80	-1.12	.265	*Pillemer & Holtzer (2015)*[63]	American older adults, community sample (*n* = 355, mean age in years= 77).	77.0 (2.50) [b]	-1.0	-3.46	**.006***
Positive Interaction	79	70.25 (3.0) 25 to 100 [b]			76.6 (25.1)	6.35	2.23	**.027***			74.75 (4.25) [b]	4.5	8.93	**.001****
Marital Satisfaction	53	19.36 (2.34) 12 to 21	*Lal & Bartle-Haring (2011)*[64]	American chronic lung disease outpatients (*n* = 52, mean age in years =66)	18.6 (3.5)	-0.76	-1.31	.193	*Herman (1994)*[65]	American older adults, community sample *n* = 168, mean age in years= 69)	18.54 (3.57)	-0.82	-1.57	.118

Note. [a] Other chronic illness groups used where PD data unavailable, [b] Scores transformed to 1-100 scale to allow for comparison.
Comparison of differences in observed mean scores for present sample with other population groups. Statistically significant differences in bold
* Correlation is significant at the 0.05 level
** Correlation is significant at the 0.01 level

Table 4

Descriptive statistics comparing SO scores on psychosocial outcomes, with scores of other PD carer and nonclinical populations.

Present Study (*n* = 58)			Other Carer Populations [a]						Non-Clinical Populations					
Psychosocial outcomes	***n***	***Mean (SD) range***	***Study***	***Sample***	***Mean (SD)***	***Mean difference***	***t***	***p***	***Study***	***Sample***	***Mean (SD)***	***Mean difference***	***t***	***p***
Psychological Health (QOL)	57	64.80 (12.22) 33.3-83.3	*Skevington & McCrate (2010)*[66]	UK elderly carers, community sample (*n* = 61)	65.78 (14.56)	0.98	0.40	.694	*Girardi Paskulin & Molzahn (2007)*[67]	Canadian older adults, community sample (*n* = 202, mean age in years = 73)	75.40 (13.26)	10.60	5.42	**.001****
Social Relationships (QOL)	56	71.06 (19.34) 16.7-100			61.68 (20.34)	-9.38	-2.56	**.012***			72.96 (17.02)	1.9	0.72	.471
Physical Health (QOL)	57	59.60 (9.37) 33.3 -78.6			61.53 (20.87)	1.93	0.5	.523			75.57 (18.7)	15.97	6.23	**.001****
Environment (QOL)	57	81.93 (12.86) 50-100			68.95 (14.67)	-12.98	-5.10	**.001****			80.18 (14.7)	-1.75	-0.82	.416
Affectionate Support	56	72.75 (4.25) 0-100 [b]	*Nightin-gale et al., (2015)*[68]	American carers of head/neck cancer (*n* = 39, mean age in years =57)	76.75 (2.75) [b]	4.0	5.17	**.001****	*Pillemer & Holtzer (2015)*[63]	American older adults, community sample (*n* = 355, mean age in years= 77)	77.0 (2.50) [b]	4.25	10.56	**.001****
Positive Interaction	57	66.25 (0.75) 0 -100 [b]			68.75 (3.0) [b]	2.5	6.04	**.001****			74.75 (4.25) [b]	8.5	15.05	**.001****
Marital Satisfaction	54	17.24 (4.13) 3 -21	*Edwards & Scheetz (2002)*[69]	American PD carers, routine care (*n* = 41, mean age in years= 67)	17.3 [c]	-	-	-	*Herman (1994)*[65]	American older adults, community sample (*n* = 168, mean age in years = 69 years)	18.54 (3.57)	1.30	2.24	**.026***
Caregiver Strain	58	3.55 (3.17) 0 -10	*Wade et al., (2003)* [53]	British PD carers, rehabilitation study (*n* = 61)	3.1 (2.7)	-0.45	-0.84	.405	-	-	-	-	-	-

Note. [a] Other carer populations used where PD carer data unavailable, [b] Scores transformed to 0-100 scale to allow for comparison, [c] SD not reported.
Comparison of differences in observed mean scores for present sample with other population groups. Statistically significant differences in bold.
* Correlation is significant at the 0.05 level
** Correlation is significant at the 0.01 level

Perceived FM Severity and Psychosocial Outcomes in PD

People with Parkinson's

Table 5 reports the correlations of PwP perceptions of FM severity with psychosocial outcomes in PwP participants. Interestingly, perceived retention of expression (expressive disposition, as measured on the BEQ) had the strongest magnitude of significant correlations with psychosocial outcomes, rather than the perceived loss of expression (FM severity as measured on the FMQ). Statistically significant correlations were found between higher BEQ scores and the following: better marital satisfaction; a higher perceived availability of affectionate/ positive social support; and fewer symptoms of depression (see table 5).

Correlations between PwP-reported FM severity and social functioning were not statistically significant. Significant, small to moderate negative correlations were found between PwP FM severity and HRQOL, and between FM severity with symptoms of depression. A significant, moderate negative correlation between FM duration and HRQOL was also found. One explanation for the latter correlation is that a longer duration of FM is related to disease progression. Consistent with this, PD stage had a significant, moderate negative correlation with HRQOL.

Some demographic and clinical variables had significant correlations with psychosocial outcomes in PwP. Small significant correlations were found between the following: female PwP gender and greater positive social interactions; shorter relationship length and more symptoms of anxiety; more advanced PD and reduced HRQOL (see table 5).

Significant Others

Correlation between SO perceived FM severity with SO psychosocial outcomes are reported in table 6. As expected, SO-reported FM severity had significant moderate correlations with relationship-related outcomes. The more FM SO perceived, the lower marital satisfaction, higher caregiver strain, and lower availability of affectionate support they experienced (table 6).

A longer duration of FM had a significant, moderate negative correlation with perceived availability of positive interactions, but was not significantly correlated with all other psychosocial outcomes measured (psychological health, general social relationships, affectionate support, marital satisfaction, and caregiver strain). There was no significant relationship between SO perceptions of FM severity and their psychological health.

Table 5

Correlations between participant characteristics with wellbeing and FM-related outcomes, for PwP (*n* = 80).

	Demographic and General PD Characteristics						FM-related Characteristics				
Psychosocial outcomes	Age in years	Gender	Relation-ship length	Age at PD Onset	H&Y Stage	Treated for Depression	FM Duration	Self-report FM Severity	Healthcare Professional rated FM Severity	Researcher rated FM Severity	Self-report Expressive Disposition
Health related Quality of Life	.03	.10	-.01	.04	**-.31****	-.10	**-.35****	**-.36****	-.04	-.19	.20
Anxiety	-.21	.16	**-.27***	-.17	.06	**.29****	.15	.03	**-.24***	-.02	.05
Depression	-.11	.03	-.14	-.11	.18	.08	.22	**.28***	-.04	.01	**-.30****
Affectionate Support	-.10	.07	.09	-.02	-.11	.21	.12	-.04	-.15	.20	**.34****
Positive Interaction	-.11	**.24***	.02	-.10	.09	.05	.06	.08	.05	.01	**.31****
Marital Satisfaction	.14	.04	.15	.15	-.05	.01	-.08	-.14	.22	-.06	**.35***

Note. Spearman's rank order correlation coefficient. Significant correlations in bold. H & Y (Hoehn & Yahr) stage is based on participant interview. Treated for Depression is self-reported pharmacological and/or psychological treatment for low mood. FM severity is the amount of perceived FM, as measured by the FMQ (PwP), MDS-UPDRS (healthcare professional evaluation), and FACES (researcher rating). Expressive disposition is PwP-reported perceived emotional expressive disposition, as measured on the BEQ.
*Correlation is significant at the 0.05 level (2-tailed)
** Correlation is significant at the 0.01 level (2-tailed)

Table 6

Correlations between participant characteristics with wellbeing and FM-related outcomes, for SO (*n* = 58)

	Demographic and General PD Characteristics				**FM-related Clinical Characteristics**				
Psychosocial outcomes	Gender	Relation-ship length	PD Duration	H &Y Stage	FM Duration	SO-report FM Severity	Healthcare Professional rated FM Severity	Researcher rated FM Severity	SO-report Expressive Disposition
Psychological Health	-.18	-.07	-.12	.15	-.01	-.17	-.10	.14	.19
Social Relationships	-.09	.16	-.12	.04	-.19	**-.28***	-.08	.09	.07
Affectionate Support	-.10	.12	.04	-.12	-.15	**-.33***	.01	.01	.24
Positive Interaction	-.16	-.04	-.09	-.15	**-.33***	-.23	.01	-.09	.22
Marital Satisfaction	**-.34***	.21	-.12	-012	-.06	**-.36****	.10	-.04	**.35****
Caregiver Strain	.15	-.23	.10	.11	.21	**.43****	.09	.10	-.21

Note. Spearman's rank order correlation coefficient. Significant correlations in bold. H & Y (Hoehn & Yahr) stage is based on participant interview. FM severity is the amount of FM perceived by rater, as measured by the FMQ (SO), MDS-UPDRS (healthcare professional evaluation), and FACES (researcher rating). Expressive disposition is SO-reported perceived emotional expressive disposition, as measured on the BEQ.

*Correlation is significant at the 0.05 level (2-tailed),

** Correlation is significant at the 0.01 level (2-tailed).

FM-Specific Difficulties and Psychosocial Outcomes in PD

Table 7 presents the correlations between FM-specific difficulty, as measured on the FMQ, with psychosocial outcomes (rather than FM symptom severity, as in the previous section). In PwP, significant small correlations were found between greater FM-related relationship difficulties and worse psychological health, and moderate significant correlations with poorer social functioning. Most notably, the more FM-related relationship difficulties PwP reported on the FMQ, the lower levels of marital satisfaction, availability of affectionate support, and availability of positive social interactions they perceived (table 7). A significant moderate correlation was also found between greater FM-related individual difficulty and more symptoms of depression. Significant, small positive correlations were found between FM-related individual difficulties and psychological health outcomes. No other significant correlations were observed between FM-related individual difficulty and psychosocial outcomes.

For SO, negative consequences of FM were most strongly correlated with outcomes related to relationship functioning. FM difficulty on the FMQ-SO had a significant moderate correlation with lower perceived affectionate support, and significant large correlations with lower marital satisfaction and greater caregiver strain (table 7). Significant, small positive correlations were also found between SO FM difficulty on the FMQ with poorer psychological health and lower levels of positive social interactions.

Table 7

Correlation between FM-related outcomes and other wellbeing outcomes in PD

Psychosocial outcomes	**FM-Specific Difficulty**	
PwP (n = 80)	**FM Relationship Difficulty Subscale**	**FM Individual Difficulty Subscale**
Health Related Quality of Life	-.19	**-.26***
Anxiety	**.25***	**.27***
Depression	**.28***	**.43****
Affectionate Support	**-.43****	-.01
Positive Interaction	**-.26***	.08
Marital Satisfaction	**-.39****	-.02
SO (n = 58)	**FM Difficulty Scale**	
Psychological Health	**-.26***	
Social Relationships	-.13	
Affectionate Support	**-.30***	
Positive Interaction	**-.27***	
Marital Satisfaction	**-.50****	
Caregiver Strain	**.53****	

Note. Spearman's rank order correlation coefficient. Significant correlations in bold.
FM-Specific Difficulty as measured on the FMQ
*Correlation is significant at the 0.05 level (2-tailed).
**Correlation is significant at the 0.01 level (2-tailed).

Other Characteristics Corresponding to FM-specific Difficulty in PD

Scores on the FMQ were also correlated with the demographic and clinical variables and are reported in table 8. The most notable findings were that healthcare professional evaluations of FM severity were not significantly correlated with any scales of FM difficulty on the FMQ, for both PwP and SO. Additionally, researcher-rated FM severity only had one significant small correlation with a single FM difficulty subscale. It was also of note that PD stage was not related to any of the FM variables.

For PwP, a younger age at PD onset had a significant moderate correlation with greater FM-related individual difficulty. Interestingly, PwP self-report of FM severity was not significantly correlated with their perception of FM-related relationship difficulties, but did have a significant, strong positive correlation with FM-related individual difficulty. For SO, female gender was the only other variable to have a statistically significant correlation with greater FM-related difficulty on the FMQ. SO-reported FM severity had a significant large positive correlation with the FM difficulty scale on the FMQ.

Table 8

Correlations between study variables and FM-specific outcomes, as measured on the FMQ.

PwP (*n* =80)		
Demographic and Clinical Characteristics	**FM Relationship Difficulty Subscale**	**FM Individual Difficulty Subscale**
Age in years	.04	**-.40****
Gender	.02	-.07
Relationship length	-.17	**-.31***
Age at PD Onset	.08	**-.42****
H&Y Stage Estimate	.09	.10
Treated for Depression	.01	.14
FM-Related Characteristics		
FM Duration	.03	**.56****
FM Severity Self-report	.10	**.63****
FM Severity Healthcare Practitioner-rated	.13	-.02
FM Severity Researcher-rated	**.24***	.11
FM Self-reported Expressive Disposition	**-.31****	.01
SO (n = 58)	**FM Difficulty Scale**	
Demographic and Clinical Characteristics		
Gender	**.29***	
Relationship length	-.17	
PD Duration	-.04	
H&Y Stage Estimate	.03	
FM-Related Characteristics		
FM Duration	.14	
FM Severity SO-report	**.53****	
FM Severity Healthcare Practitioner-rated	.04	
FM Severity Researcher-rated	.20	
FM SO-reported Expressive Disposition	**-.58****	

Note. Spearman rank order correlation coefficient. Significant correlations in bold. H & Y (Hoehn & Yahr) stage is based on participant interview. FM severity is the amount of FM perceived by rater, as measured by the FMQ (PwP/SO), MDS-UPDRS (healthcare professional evaluation), and FACES (researcher rating). FM Knowledge is the level of symptom knowledge, as reported the FMQ.
*Correlation is significant at the 0.05 level (2-tailed),
** Correlation is significant at the 0.01 level (2-tailed).

Discussion

This paper examined the association between FM and psychosocial outcomes in PwP (people who have PD) and their SO (spouse/partner or other close family member). The primary aim was to explore the psychosocial correlates of having FM. The secondary aim was to compare how different observers perceived FM.

Overall, greater SO perceptions of FM severity and lower PwP perceptions of retained expressivity were the variables most strongly associated with greater FM-related difficulty, which in turn was the study variable most significantly related to poorer psychosocial outcomes. For PwP, the more negative consequences of FM that were reported, the more the anxiety and depression symptoms that they experienced. They also experienced lower levels of marital satisfaction, perceived a lower availability of affectionate support and a lower availability of positive social interactions. For SO, the more negative consequences of FM that were reported, the lower the level of marital satisfaction, more caregiver strain, the less available they perceived affectionate support or positive social interaction, and the worse psychological health they experienced. These findings share similarities with previous evidence exploring the associations between FM and reduced social functioning in PD caregivers [31] and compromised quality of life in PwP who have greater FM [32]. Given the limited number of current studies, further investigation is required to clarify our understanding of the psychosocial consequences this symptom.

It was noteworthy that PwP did not report compromised social relationship functioning when compared to other PD populations and to normative samples of older adults, but SO did**.** This may occur because the person with PD's perception of the relationship is protected, possibly by disease-related deficits in their own emotion recognition abilities, as suggested by Gunnery and colleagues [31]. It could also be that FM affects the social functioning of significant others differently to people who have PD, because significant others face the challenge of balancing caring for a person with PD and keeping up with changing roles in the relationship and in the responsibilities of everyday life. The good

overall social functioning reported by participants with PD may also be one factor to impact favourably on their psychological health, which represented normal (nonclinical) levels of psychological distress, and surprisingly, was found to be statistically better than that reported in other PD populations and similarly aged adults. An additional explanation for this may be that participants self-selected based on a relatively low levels of distress and other psychosocial stressors. Notwithstanding this limitation, we found greater FM-related difficulty was significantly correlated with more symptoms of depression and anxiety for PwP, and to poorer psychological health and decreased social functioning in SO.

Only a few demographic and general clinical variables were found to correspond with greater FM-related difficulty. What stood out was the lack of significant correlations between PD stage and most psychosocial outcomes measured. This may suggest that the effects of FM are somewhat independent of PD progression. We also found the effects of FM were most marked for younger PwP. It is possible this reflects the impact of having a progressive condition at a life stage where there is greater expectation of health and higher responsibilities in work and family roles.

Differences were found in raters perceptions of FM severity. We also found that associations between perceptions of FM and psychosocial outcomes/FM difficulty were not consistent across raters. The most striking finding was that healthcare professional evaluations of FM severity were largely unrelated to most psychosocial outcomes that were measured, and unrelated to reports of FM-specific difficulties, when compared to PwP/SO ratings of FM. These findings are congruent with the idea that observations of clinicians differ from the perspectives of people who live with FM every day [32] and are consistent with findings showing different social relationships influence the amount of expressivity displayed by people who have PD [11]. There is a need for investigation of how healthcare professional ratings of FM staging done in routine clinical settings relate to impressions of individuals with FM in primary social relationships, and relate to the psychosocial correlates

of FM. For example, how does the impression a PwP is grumpy all time relate to the presence and degree of clinical markers of FM typically evaluated in the MDS-UPDRS [23].

It should be noted that these differences may be due in part to the different ways of assessing FM. The videoed discussion may not approximate the level of expressivity displayed in everyday family life or in usual interactions during routine PD care. Assessment by treating healthcare professionals, who are more familiar with the individual with PD, may agree better with PwP/SO FM ratings. That said, the present findings suggest that clinical ratings of FM do not provide sufficient information to evaluate the emotional and social consequences of FM, or to identify who experiences the most distress relating to this symptom. The findings further suggest it is important to ask PwP/SO about their experiences of FM, because their perceptions of FM difficulty were found to relate most strongly to wider psychosocial problems.

The limitations of this exploratory study are the use of mainly correlational analyses and participant recruitment from a community sample where participants may have self-selected based on relatively good psychological health and/or an earlier stage of PD. Further confirmatory research is needed to investigate how FM-specific difficulties may lead to poorer psychosocial outcomes in PD and to develop interventions to lessen the impact of FM. Future studies could be improved by using the same assessment methods or instruments across expert, family, and patient groups. Other variables that could be usefully explored which relate to the clinical evaluation and management of FM may include the following: adjustment to having FM including concepts such as psychological flexibility; fluctuations in FM (state-trait-like dimensions of FM severity); variations in FM experience based on sociocultural variables such as gender; a possible interaction between PD stage (and/or FM severity) and the ability to meet social role demands across the stages of life; the potential benefits of education in increasing understanding of FM; and the usefulness of various compensatory strategies. It is also noted that unequal gender distributions in PwP and SO groups of the present study could have influenced the findings. Other disease-

related changes imposed by PD may also have influenced the way respondents answered the FMQ items. Future investigations could usually include a larger sample of female participants with PD/FM and include clinical measurement of PD staging.

Conclusion

We identified that FM is related to poorer psychosocial outcomes in PD, however PwP and SO may be affected in different ways. The findings also highlight the importance of including patient and significant other perspectives of FM. Clinical evaluation of FM provides useful information about the staging of this symptom, but PwP/SO experiences of FM should be included in addressing the consequences of this symptom in the everyday lives of people who have PD and their families.

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Supplementary Data

Study 3 Appendix I. Rater Instruction Guide for Coding of Video Data

Three main rating variables are used in this research to measure facial expressivity are: expression; neutral; and masked neutral. The Facial Expression Coding System (FACES) was originally developed for measuring the frequency, duration, valence & intensity of facial expressions (Kring & Sloan, 2007). In this research, the FACES has been adapted for use with a Parkinson's population who may experience bradykinesia (slowness of movement), akinesia (poverty of spontaneous movement) and hypokinesia (reduced amplitude of movement) in the musculature of the body and the face. The results in deficits in the expression of emotion, called hypomimia or facial masking.

Please follow the original FACES user guidelines to code the frequency, duration, valence, and intensity of emotional expressions (Kring & Sloan, 1991, The facial expression coding system: A users guide. Unpublished manuscript. pp. 2-8). To capture the nature of expressive dysfunction in Parkinson's, this research introduces additional variables (or codes) to the FACES. This includes a 'Neutral' code which measures the frequency and duration of neutral facial displays. To measure the unique characteristics of hypomimia, an exploratory 'Masked Neutral' code was created. This represents the lack of expression plus other clinical features of common to people with hypomimia/facial masking. Additionally, the proportion of talking/not talking of each video clip will be measured. The following sections detail these additional codes and other things to note when observing individuals who have Parkinson's.

Detecting Neutral Displays – Additional to original FACES

This is the neutral configuration of the face when there is no observable use of facial muscles. There is no observable facial activity or repositioning of the facial muscles. Neutral looks slightly different on each individual- it may not always look the same in each instance it is observed. Coding neutral will become easier as you develop a familiarity with the individual.

Take care to differentiate neutral from a low intensity expression. For example, when an individual is listening or thinking, they may appear neutral or may display an expression like concentration at a low level. If there is expressive muscle movement or clear affect can be observed, the coder should instead record an expression,.

Detecting Masked Neutral – Additional to original FACES

This is a somewhat experimental code. It records characteristic features of Parkinson's facial masking, in addition to a lack of expression. It may be useful to think of this code as Neutral *plus* features of facial masking. This is defined as an at-rest facial configuration *plus* one or more of the following:

i. Reduced blink frequency (staring).

ii. Unintentional lip separation (mouth open) or lips closed with jaw dropped and unintentional downturned lip corners.

iii. Unusually fixed (static) eyebrows which may be raised or furrowed.

iv. A "stone-faced" rather than neutral or at rest appearance.

The general impression or demeanour is often negative, rather than at rest, while still not showing any active expressivity. For example, cold, uncaring, unfriendly, aloof, disinterested, inattentive, bored, grumpy, zoned out or cognitively impaired.

It can be difficult to determine this code from neutral at first. Make sure to attend to the overt (easily visible) presence of any masking features (mentioned above). If unsure, a neutral code is likely more appropriate. Note that if a participant opens their mouth to breath

or sigh, this is not counted as masked neutral.

It may also be possible to see features of this code combined with expression or gesture in other areas of the face. For example, a staring appearance and fixed upper face might be accompanied by a smile or grimace in the lower face. Since codes are mutually exclusive, meaning only one can be coded at a time, any observed expression takes precedence over other code types.

Masked Neutral looks slightly different on each individual. As you become more familiar with each person you will find it easier to identify and discriminate how masked neutral/neutral appears for that individual.

Other Things to Note:

Parkinson's Symptoms

Participants may display facial or bodily movements related to their condition. These movements can be mistaken for expressions of emotion and/or make it challenging to view facial movement. Symptoms commonly observed in the participants of this study are listed below. If a distinct physical symptom is observed, carefully attend to other areas of movement in the face to discern if a facial behaviour can be coded or not.

Symptom	Definition	Exemplar
Tremor	Shaking, quivering or repetitive rhythmic movement. Characteristically occurs when at rest.	From mild trembling of the jaw, a head 'bobble', to intense shaking of extremities or the entire body. This can appear to be distress, so examine other areas of facial movements to determine an expression.
Dyskinesia	Uncontrolled movement associated with use of anti-Parkinson's medications.	Writhing, twisting or twitching movements. May be one body part or the entire body. May occlude view of the face.
Dystonia	Cramping, spasm or twisting of muscles.	Usually, one or two areas of the face will appear to twist or contract. This

		symptom can be frustrating and painful. Attend to other areas of facial movement and code an emotional expression if present.
Postural Changes	Awkward, sustained positioning of the head or trunk	Hunching over, stooping, dropped shoulders or head, leaning forward. A few participants seem to do the opposite by angling their head upwards and away, in a manner that can appear aloof. May occlude view of the face.
Reduced Blink Frequency	Because there is less lubrication eyes become teary or watery.	Can appear to be weeping. Easily mistaken for sadness when combined with a jaw tremor. Examine other areas of facial movement to determine whether an expression can be coded.
Hypomimia	The reduction of spontaneous and voluntary movement of muscles of the face.	A fixed or stone-like appearance, reduced blink frequency (staring), unintentional lip separation (mouth open) or unintentional downturned lip corners, fixed eyebrows which may be raised or furrowed. Gestalt impression is often negative rather than simply at rest. Code this as Masked Neutral.

Please respect the privacy of study participants by not discussing the individuals you observe outside the viewing room.

Study 3 Appendix II. FM Compensatory Behaviours and Coping Strategies

Tables 1 and 2 summarise the types of compensatory behaviours and coping strategies used by PwP and SO, as reported on open answer items of the FMQ.

Table 1

Strategies reported by PwP that help them cope with FM

Compensatory Behaviours and Coping Strategies	Quotes from open answer item of the FMQ
Exercising the face	"When my face feels fixed I do exercises that stretch and move my face. It helps to a degree. It eases the contracted, fixed feeling of the muscles. "Sometimes I pull faces in the mirror when cleaning my teeth or having a shower". 'I use my face more at gym because it's hard physical work"
Group Rehabilitation Programme	"I attend [group] singers where facial expression exercises are given with the vocal exercises". "I believe that LSVT voice training has helped maintain my facial mobility".
Posing expressions and using bodily gesture	"Try to smile when I think it's appropriate. For example, if I'm feeling happy or someone has said something amusing, or when I've done something well. I wouldn't smile unless I've thought about it. This is helpful". "When meeting friends or acquaintances, if stopping to talk to them, I try to force a smile. As if taking a photo and you try to smile. Being conscious of it makes it look better. But the impression you get from people is that you're still not all there sort of thing".
Humour	"I use whatever I can to keep engagement with others. Other tactics for example, like humour when public speaking or interacting with others".
Educating Others	"I give people a poster that I made about FM, and I tell them about it (when in NZ). People at church here, when they see me, have a query. Now they don't worry about it as much. In Asia, I didn't say anything about FM. But when I question them, they say I look okay. But I don't think what they told is 100%. I do think it's helpful to mention it to people that you know. For example, say, I may look this way, but I don't feel like that".
Positive and Accepting Outlook	"I accept it and keep going out with people. Don't worry about how you look and feel I tell myself sometimes. But I do limit myself sometimes". "I have a picture in my mind where I am happy and smiling. I believe I reflect that in my face"
Taking extra medication	"I extra take Parkinson medication when socializing or going out. Shaking seems to reduce but I'm not sure about the masking, but I think it is also reduced".
Being sensitive to others and deliberate in interactions	"I try to listen to people and understand their situation. This helps". "With new people I try to be the best I can be" "Kiss and cuddle makes me happy"

Table 2

Strategies reported by SO that help them cope with FM

Compensatory Behaviours and Coping Strategies	Quotes from open answer item of the FMQ
Asking questions and agreeing to check in	"I used to ask him if he was alright and he got angry with me asking all the time. So now I just don't say anything on the agreement if something is wrong he will tell me". "Although I have felt before my dad was diagnosed that his expressions can be hard to interpret, there has seemed to be a slight change since his diagnosis. Sometimes I am unsure how exactly to reconcile his expression with his words, so often I will ask further questions to help determine his mood". "I sometimes ask directly how he is feeling, does he want to do 'whatever', is he happy with what we are doing, what we have planned - so he can tell me even if he can't show me by expression. This works well assuming he is honest!"
Creating awareness of FM	"I tell him when he looks grumpy" "Reminding him to smile sometimes helps" "I tell him what he is displaying that I find difficult. He makes an attempt to change this expression"
Relying on other cues	"Focus on body language instead" "I try to sit directly in front of him so I can see his eyes/eye movements, and to draw his face to mine"
Catching out misinterpretations of FM	"I remind myself that [her] masking is not a reflection of her emotional state, it is neither intentional or a reflection of her mood." "Just keep trying to remember how he looks (in himself or what he is seeing) is not related to how he is feeling, (in himself, about himself, or about me or life in general)"
Humour	"We sometimes poke fun at each other and crack each other up"
Avoidance	"In some cases I simply look elsewhere to avoid being dismayed by a slack jaw look or staring eyes (especially when watching tv)"
Educating others	"I have used and distributed a Parkinson's fact sheet on facial masking to family and friends, so they have more understanding of [my husband's] responsiveness. This was very helpful". * "Have to tell people about his Parkinson's so they understand his behaviour"
Positive and accepting outlook	"I understand that is part of the condition and accept that it is involuntary". "I tell myself that it is not how it he wants to be but how the disease is affecting him." "Not dwelling on it" "Each of the changes i.e., facial masking etc is a challenge to be understood and worked through" "I tend to treasure the times when I do see her smile even more."
Seeking professional help	"After seeing a psychotherapist, I learnt ways to disengage from my concerns/worries".
Being patient and deliberate in interactions together	"We try to smile and laugh a lot together" "We are working on creating other areas and ways to express our closeness just being together." "Engaging him in conversation will usually enliven his face"

*Fact sheet developed during this research, see thesis appendices.

7. Integrated Findings Summary

To evaluate the consistency between findings of the qualitative investigation and the quantitative investigation, the mixed-method inferences of this research project are described in a series of joint displays and brief summaries. Qualitative findings came from all PwP and their SO's who were interviewed in Study 1. Quantitative findings comprise the results of Study 2 and Study 3. FMQ item response distributions reported here refer to the subsample of PwP/SO who self-reported FM, unless otherwise stated.

A joint display summarising the findings of how different observers perceived FM is provided in Figure 3 (see page 176). The lack of statistically significant correlations between PwP/SO ratings of FM severity with healthcare professional ratings was a striking finding. Further comparisons, for example of the concordance of ratings of the degree of FM severity, were limited because different methods and measurement tools were used across the rater groups. Comparisons of the frequency of FM/no FM ratings across the four rater groups, however, suggested the self-reports of the PwP may underestimate FM severity when it is a lower level.

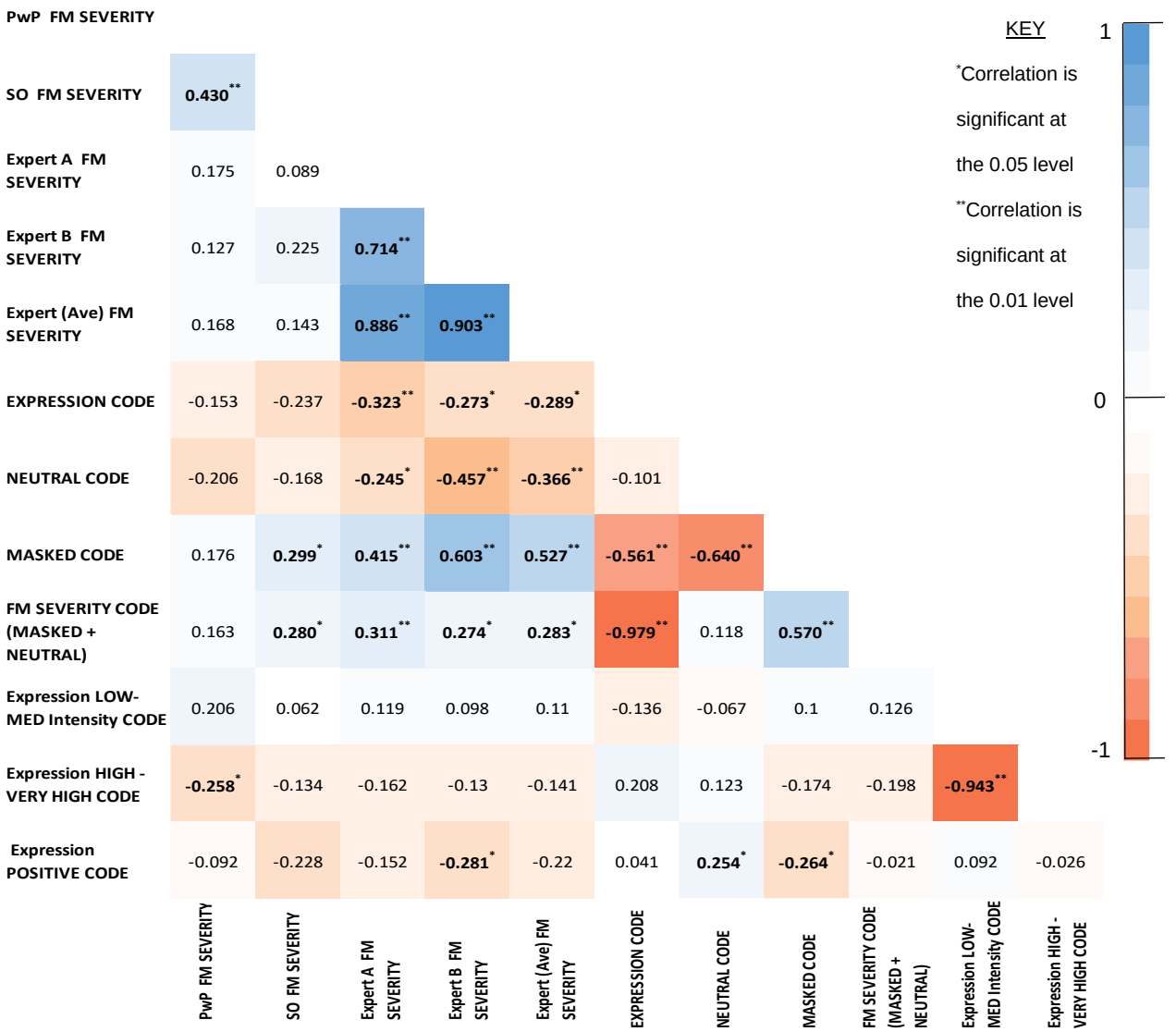
When comparing attributes of FM emphasised in personal accounts with researcher-rated facial expressive behaviours based on the videoed conversation, there was a mix of convergence and divergence of findings. Less frequent smiling was commonly mentioned as characteristic of FM in the interview findings, but a relationship was not found between the proportion of positive smiles in the videotaped samples and PwP/SO perceptions of FM severity. A lower amount of overall expressivity was also emphasised in the interviews, with decreased instances of expression in the videos found to be significantly related to perceptions of FM severity for PwP and SO. Another clinical feature of FM that was prominent in the interview findings was an appearance of blankly staring. A reduced blink frequency (which gives the appearance of staring) as part of the "masked" facial behaviour code from researcher ratings was found to be related to perceived FM severity ratings of

healthcare professionals and SOs, but not to PwP ratings of FM severity. Lastly, people who took part in the interviews also reported their or their partner's expressions to be “watered down” or reduced in amplitude. However, a lower intensity of observed expressions in the researcher-rated video data was not related with ratings of FM severity for PwP, SOs, or healthcare professionals. Changes to the speed or onset of expressions, as reported in the qualitative data, was unable to be quantitatively assessed. Respondent feedback collected about the FMQ extended the interview findings, as it revealed reports that fluctuations in the level of FM occurred. Some participants stated that instances of relatively normal expression were possible, even if only rarely.

Figure 3

Joint Display Integrating the Qualitative and Quantitative findings on perceived expressivity by rater

QUALITATIVE FINDINGS *(n = 9 PwP, n = 9 SO)*	QUANTITATIVE FINDINGS *(n = 74 PwP, n = 54 SO, consenting to video sample)*	SUMMARY OF MIXED METHOD INFERENCES
THEME Disturbances of expression ***SUBTHEME OR CODE (S)*** Expressions reduced in size, frequency, & delayed onset. Loss of or distorted smiling Blank & staring appearance	**Correlation matrix showing strength of relationships between perceived FM severity, by rater type.**	Overall, a dissimilar presence of FM was reported across rater groups. Changes in FM severity ratings were not strongly correlated between rater groups. As expected, PwP and SO pairs had moderate positive association. Attributes of FM emphasized in personal accounts, converged with some findings from researcher-rated facial expressive behaviours observed in the videotaped conversations, but not others.



Findings about the social discomfort associated with having a facial difference were not strongly consistent (see Figure 4, pg. 178). PwP interviewees described having unpleasant experiences during social interactions due to FM. This included with close friends or family members, as well as in more casual interactions. FMQ items response were not strongly consistent with this finding. A minority of PwP endorsed items relating to feelings of awkwardness and avoidance in their close relationships, and very few endorsed the item that others had been unkind about their facial appearance, those that did reported it was an infrequent experience. Responses to open answer FMQ items extended the findings relating to facial difference further, for example:

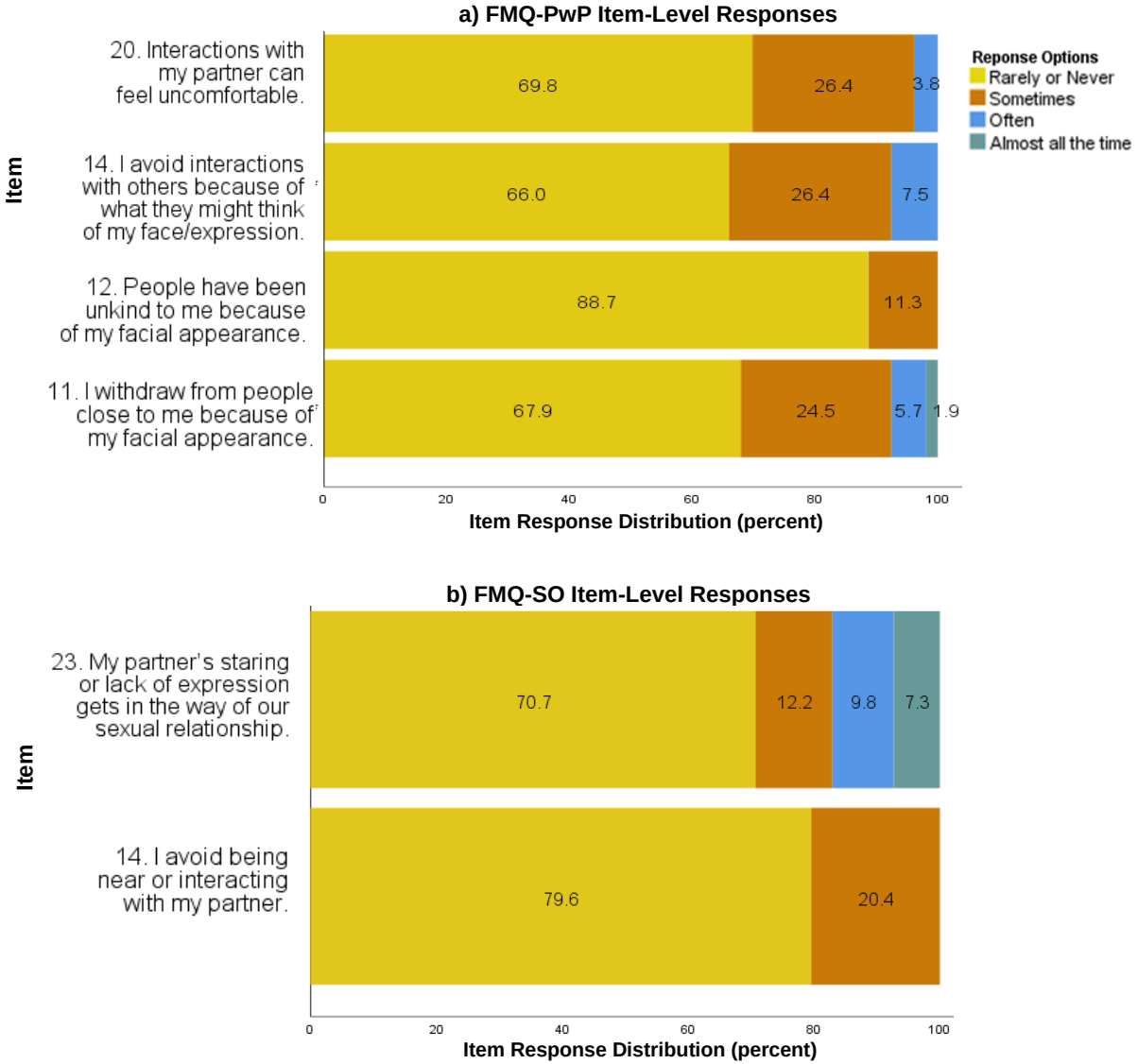
> *I'm more self-conscious about facial masking than other symptoms. You can hide your shakes... But the facial masking thing, people say, 'what's wrong with him?'…If I had to choose between a reduction in physical mobility and facial masking and speech, I would rather end up in a wheelchair and still be able to communicate. It's a higher priority for me as a person than my mobility… Communicating using my face is also important so that I can continue doing my job and contributing to the world*.

Fear of being evaluated negatively due to FM, or intense anxiety about the future consequences of FM, were not such strong thematic elements in the original interviews and therefore were not included in FMQ items.

SO interviewees described a sense of unease due to the lack of expressive response, which could negatively affect their sexual relationship. They also described a general reluctance or avoidance of interacting with the PwP. About one third of SO reported FM was an obstacle to physical intimacy, with around half of those responding it was a frequent experience. FMQ responses indicated around 20% of SO reported avoidance of interacting with the PwP, although most SO endorsed this as an infrequent experience.

Figure 4

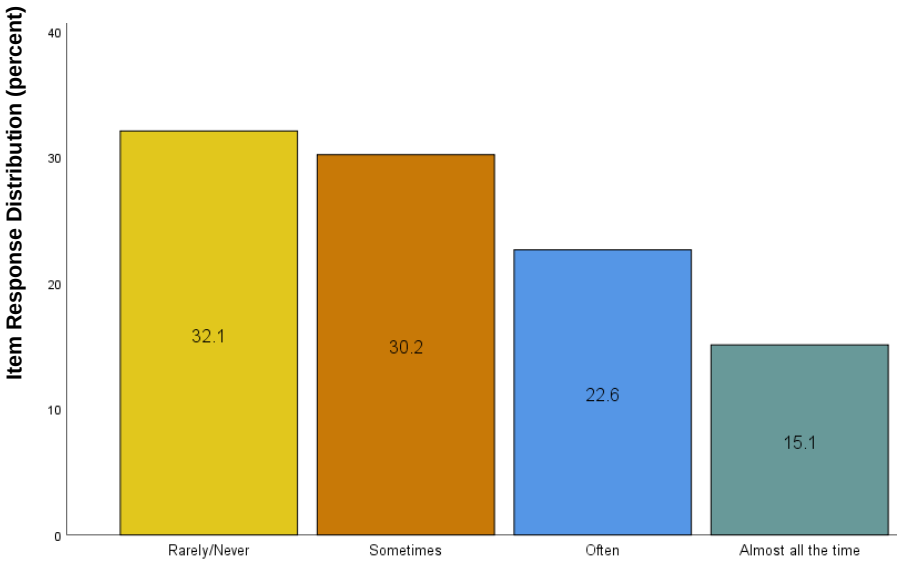
Joint Display Integrating the Qualitative and Quantitative Findings on the intrapersonal and interpersonal discomfort of having FM.

QUALITATIVE FINDINGS (*n = 9 PwP, n = 9 SO*)	**QUANTITATIVE FINDINGS** *(n = 53 PwP, n = 50 SO, subsample self-reporting FM)*	**SUMMARY OF MIXED METHOD INFERENCES**
THEME Discomfort of facial difference ***SUBTHEME OR CODE (S)*** Avoidance or reluctance to interact. Sense of unease (sexual intimacy) Unpleasant reactions (casual interactions)		Interview findings showed that having FM could be an unpleasant experience for PwP during social interactions with close friends or family, as well as with casual acquaintances or strangers. On the FMQ, around one quarter of PwP respondents endorsed items relating to these themes, although most rated these as relatively infrequent experiences. For SO, it was partially confirmed that a sense of unease and avoidance could result from FM. FMQ item responses showed around one third of SO felt FM was an obstacle to physical intimacy. Findings were less consistent about SO avoidance of the PwP, with around one fifth of SO endorsing this item an infrequent experience.

Most PwP found posing socially appropriate voluntary expressions unfavourable as a strategy to compensate for the loss of spontaneous emotional expression (see Figure 5). Interviewees commented it was possible to form posed expressions, but when they did it felt inauthentic and/or required a great deal of mental and physical effort. Over half of the FMQ respondents self-reporting FM had a similar experience when making a posed expression. When PwP were asked to list what they found helpful to cope with reduced expression, around 15% of those self-reporting FM (*n* = 8) said they attempted to make appropriate expressions on occasion, such as a smile. It was unable to be assessed whether smiles were genuine or not in the video data.

Figure 5

Joint display integrating the Qualitative and Quantitative findings on posed expressions feeling effortful and inauthentic.

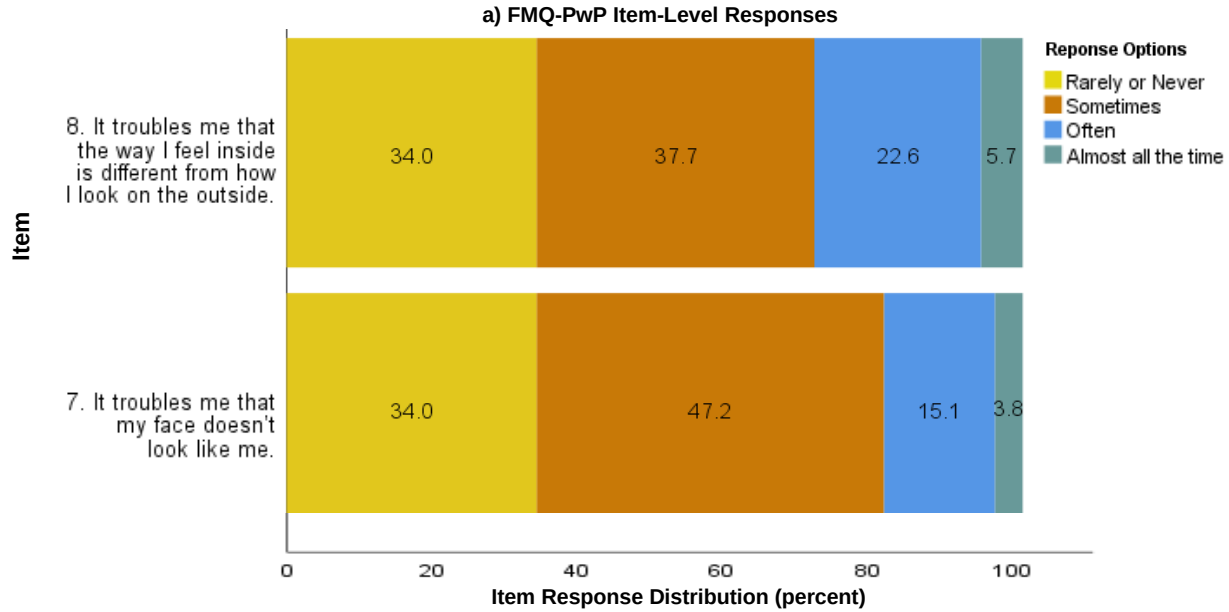
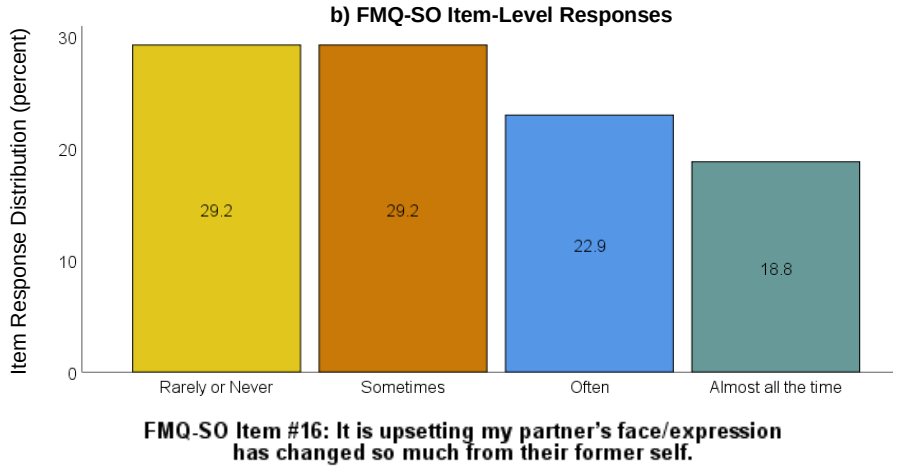
QUALITATIVE FINDINGS *(n = 9 PwP)*	**QUANTITATIVE FINDINGS** *(n = 53 PwP, subsample self-reporting FM)*	**SUMMARY OF MIXED METHOD INFERENCES**
THEME Inauthentic & effortful voluntary expressions	**FMQ-PwP Item-Level Responses**  **Response Options (Item #5: When I try to make an expression, like a smile, it takes a lot of effort, feels false, or "put on").**	It was confirmed that PwP found posing voluntary expressions (to substitute the loss of spontaneous facial expression) contrived and effortful.

PwP Interviewees felt a sense of “facelessness” where they were losing their identity and bodily autonomy, which was linked to FM. This was consistent with results on the FMQ, where just over half of PwP endorsed FMQ items relating to an experience of distress at their decreased ability to outwardly display they felt they were person inside (see figure 6, pg. 181). Additionally, in Study 3 a significant positive correlation was found between FM and psychological distress, with the greater degree of FM-related difficulty PwP described, the more symptoms of depression and anxiety they reported (see Chapter 6).

Findings that SO experienced distress at not being able to see their loved one in the masked face were consistent in the quantitative and qualitative findings also converged. A large proportion of SO endorsed FMQ items relating to emotional distress and strain caused by disease-related changes in facial appearance. For SO in Study 3, FM-related difficulty had a significant, moderate, positive correlation with caregiver strain (see Chapter 6).

Figure 6

Joint Display Integrating the Qualitative and Quantitative findings on the experience of "facelessness".

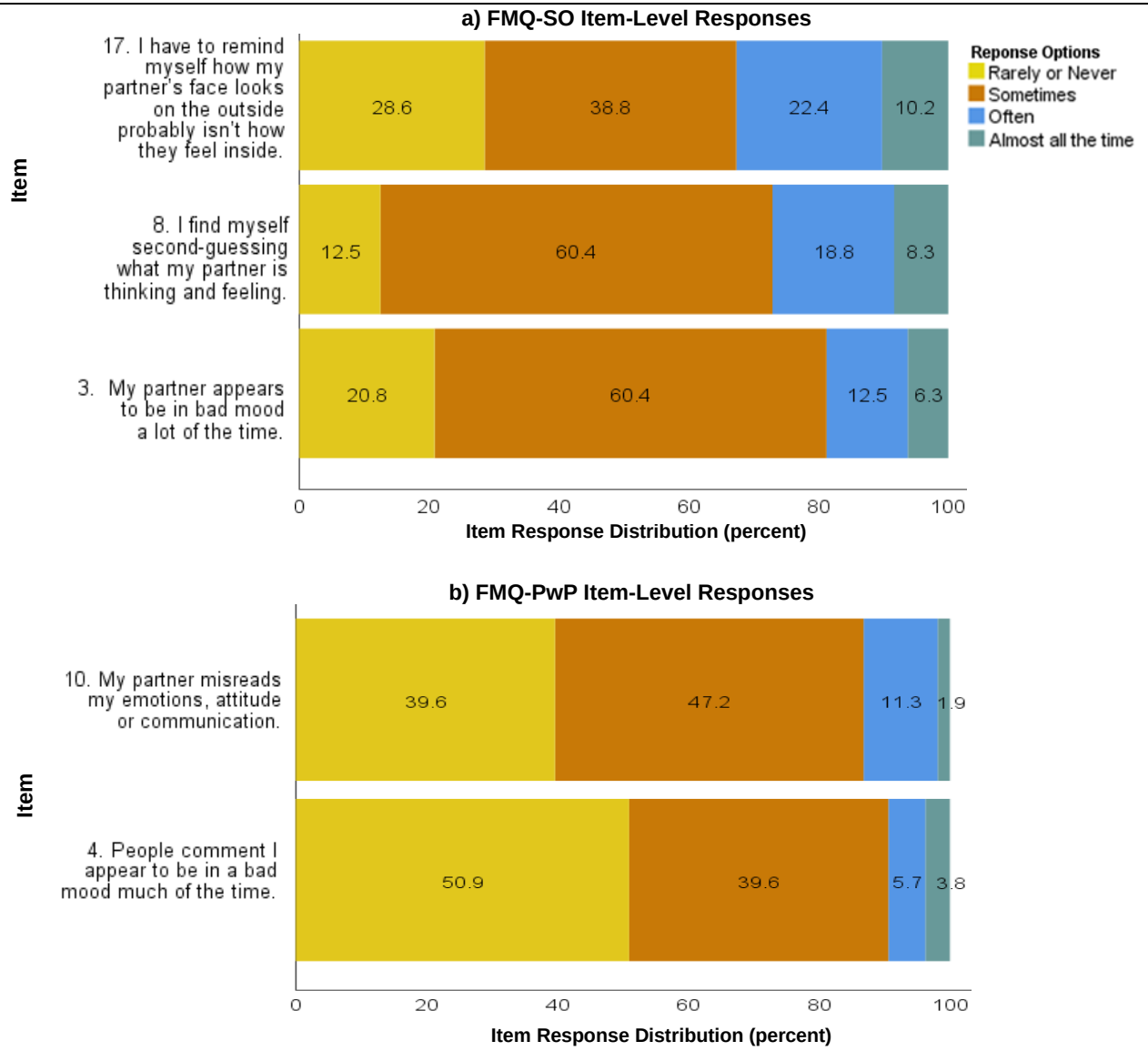
QUALITATIVE FINDINGS *(n = 9 PwP, n = 9 SO)*	**QUANTITATIVE FINDINGS** *(n = 53 PwP, n = 50 SO, subsample self-reporting FM)*	**SUMMARY OF MIXED METHOD INFERENCES**
THEME Facelessness ***SUBTHEME & CODE(S)*** Mismatch of internal experience and outward expression Unable to fully embody (express) internal experience Loss of bodily autonomy Self-concept challenged Shared identity challenged	 	The findings converged regarding the experience of "facelessness"- the feeling that oneself or one's partner was being lost. It was also confirmed that SO experienced distress at not being able see the person beneath the masked face, with most SO endorsing FMQ items relating to experiences of distress resulting from changes to the facial appearance of the PwP.

Findings about the misidentification of FM as a negative affect converged. A joint display summarising the findings is provided in Figure 7 (see pg. 183). SO interviewees described that FM could lead to misconceptions the PwP was persistently in a negative affective state, such as sad, angry, bored, or disinterested. FMQ item response distributions to relevant questions were consistent with this finding. The majority of SO responded they misinterpreted FM as a bad mood, and many experienced this frequently. PwP responses to items relating to misreading FM converged, with most respondents indicating they perceived others read their emotion, attitude, or communication inaccurately.

Some SO interviewees described a state of uncertainty associated with the lack of clear cues of emotion or intention, with other SO reporting they automatically misread FM and had to remind themselves they could not rely on facial expressions (or lack thereof) despite having knowledge of FM in PD. SO responses to FMQ related items confirmed these findings. The vast majority of SO endorsed having to backtrack and remind themselves of this cognitive bias and second-guessing what the PwP was thinking or feeling. These experiences were reported to occur highly frequently for SO.

Figure 7

Joint Display Integrating the Qualitative and Quantitative findings on the negative impression bias resulting from FM.

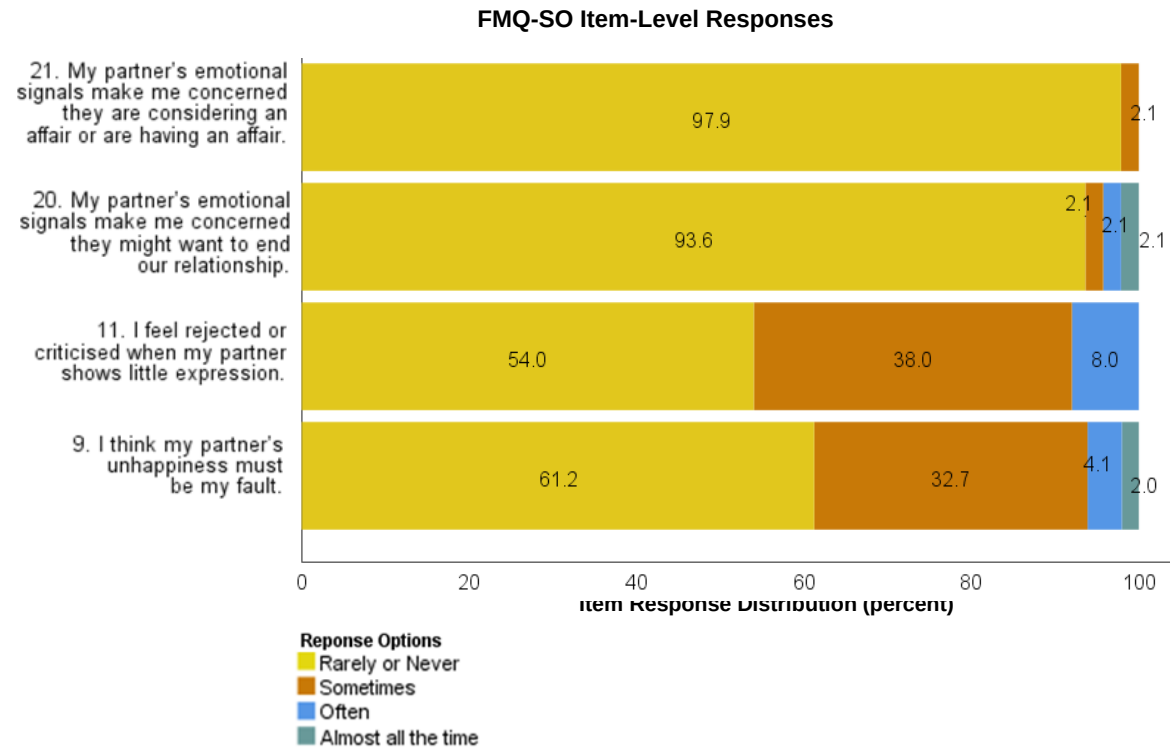
QUALITATIVE FINDINGS *(n = 9 PwP, n = 9 SO)*	QUANTITATIVE FINDINGS *(n = 53 PwP, n = 50 SO, subsample self-reporting FM)*	SUMMARY OF MIXED METHOD INFERENCES
THEME Misidentification of FM as negative affect **SUBTHEME & CODE (S)** Backtracking and second guessing		The finding that SO commonly perceived FM as negative affect was confirmed. It was also confirmed that most PwP experienced their facial behaviour to be misread, even in perceivers who were close to them.

Findings that SO often attributed incorrect reasons for the lack of expression converged. A joint display summarising the findings is provided in Figure 8 (pg. 185). SO interviewees reported that they misread PwP emotion, attitude, or intention, and based on these judgements could make assumptions that were negative and self-blaming. FMQ item response distributions were consistent with these findings. Around half of SO endorsed FMQ items that they blamed themselves for the apparent bad mood of the PwP, and just under half of SO reported feeling hurt and rejected at FM.

A small number of SO interviewees described how FM could lead to a loss of confidence in the relationship or concerns about fidelity. SO found these experiences distressing and confusing because they did not reflect the nature of their relationship history with the person who had PD. FMQ item response distributions partly converged with this finding, indicating a handful of SO's experienced concerns about the relationships, and for one SO, concern about fidelity of the PwP.

Figure 8

Joint Display Integrating the Qualitative and Quantitative findings on the misattribution of perceived lack of expression.

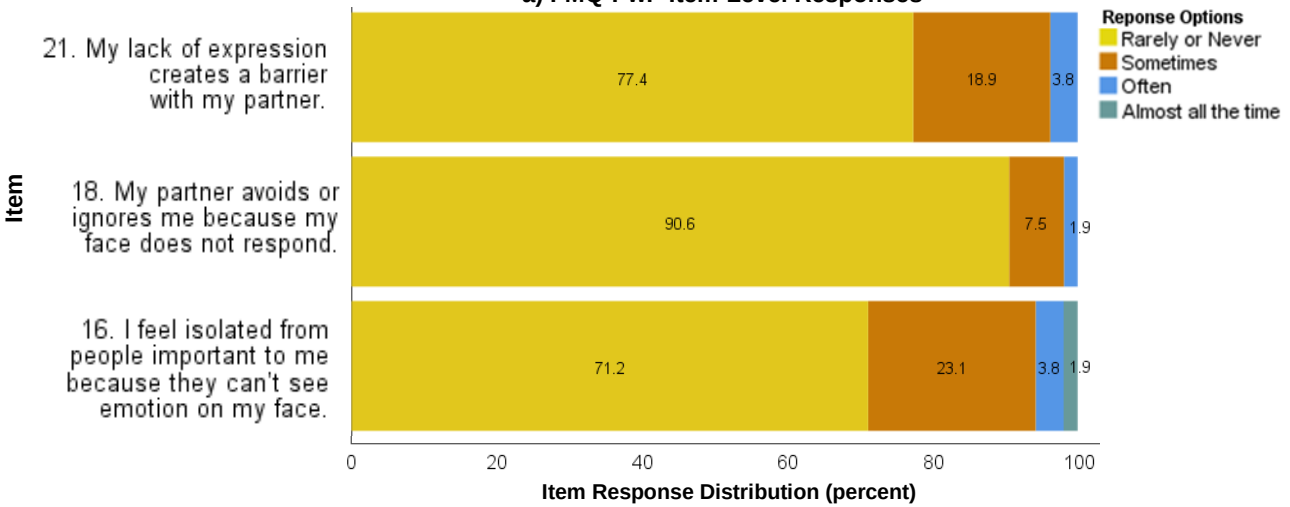
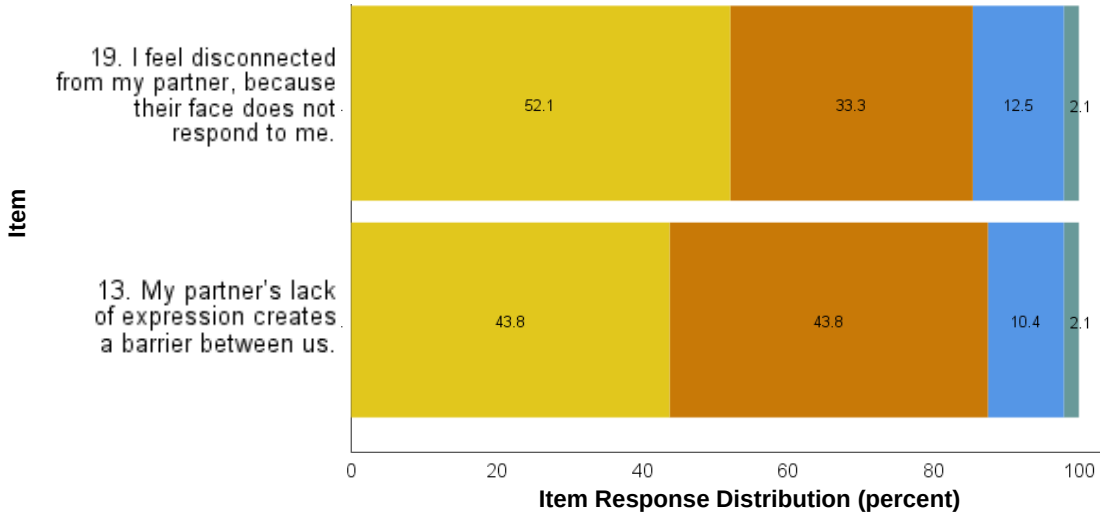
QUALITATIVE FINDINGS *(n = 9 SO)*	**QUANTITATIVE FINDINGS** *(n = 50 SO, subsample self-reporting FM)*	**SUMMARY OF MIXED METHOD INFERENCES**
THEME (Mis)attribution of FM **SUBTHEME & CODE (S)** Internally directed negative assumptions Questioning commitment or concerns about trust		SO interviewees described misattributing the reason for the lack of expression, and that their assumptions about the reasons for the PwP appearance tended to be negative and self-blaming. Overall, SO responses to items on the FMQ were consistent with these findings. A small number of SO interviewees described how FM could lead to doubts about commitment or trust in their relationship. FMQ item responses were consistent with this finding, indicating these concerns were mostly infrequent experiences reported by a small handful of SO.

A sense of growing disconnection, which was found in the interview themes, was partially consistent with the overall quantitative findings. In the interviews, some PwP described FM as a “emotional stonewall” between them and their family members, and that it can “put a couple apart”. FMQ items relevant to this theme were not endorsed as frequently or highly as expected by the subset of PwP reporting FM, given the importance and relevance of these themes for some interviewees (see figure 9, pg. 187). However, in Study 3 (*n* = 80 PwP), greater levels of overall FM-related difficulty were found to have small significant associations to some aspects of social relationship functioning measured, such as marital satisfaction and perceived availability of affectionate support (see Chapter 6).

On the other hand, findings of a sense of emotional disconnection converged for SO. Some SO interview participants described FM as an interpersonal “gulf” between them and the PwP. FMQ Item response distributions of the subset of SO reporting FM indicated around half felt a sense of disconnection or alienation in response to the lack of expression. This was also consistent with the findings of Study 3 (n = 58 SO), where increased FM-related difficulties had moderate sigficant correlations with poorer social relationship outcomes, such as reduced marital satisfaction and a lower perceived availability of affectionate support (see Chapter 6).

Figure 9

Joint Display Integrating the Qualitative and Quantitative findings on the sense of alienation and emotional detachment linked to facial masking

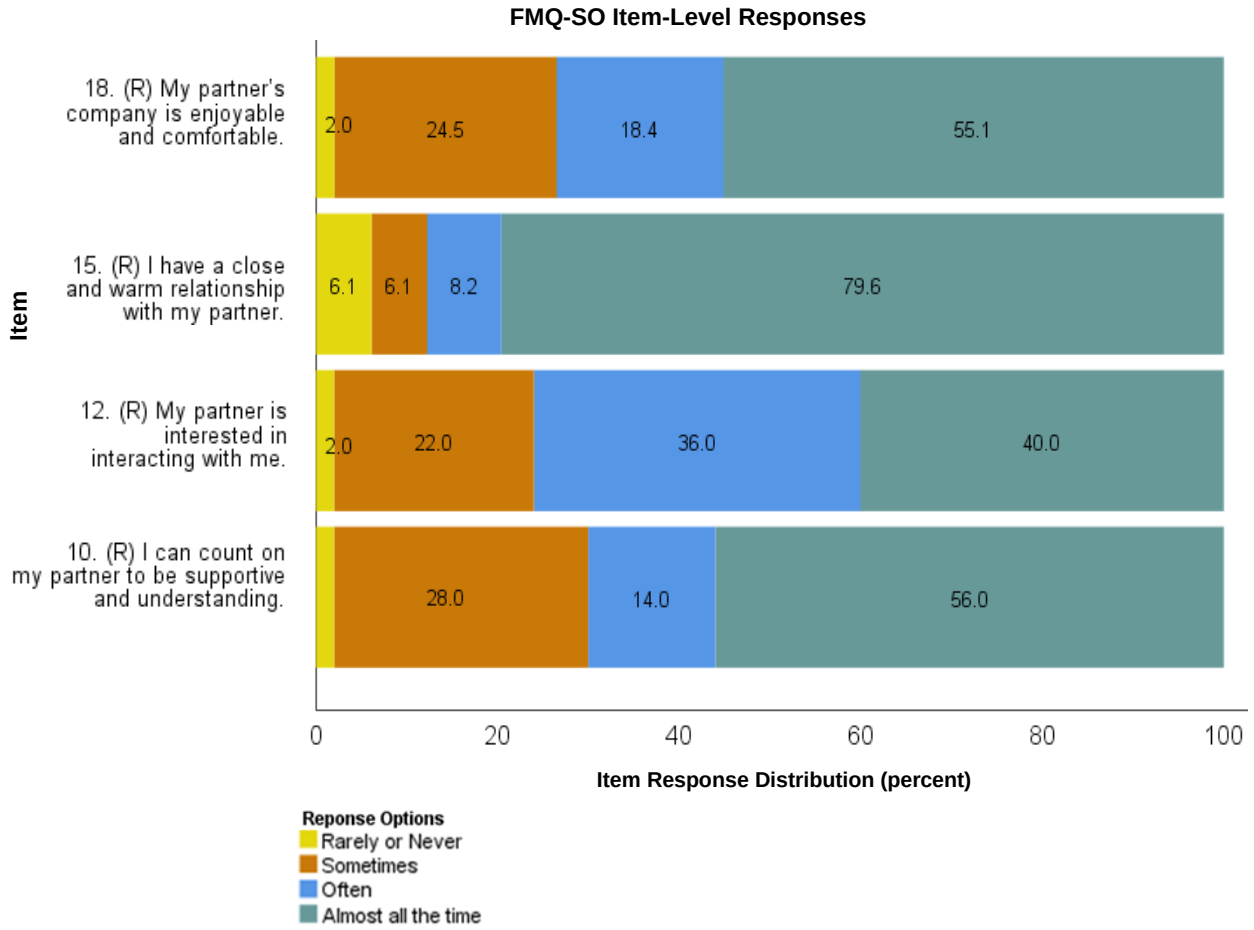
QUALITATIVE FINDINGS *(n = 9 PwP, n = 9 SO)*	**QUANTITATIVE FINDINGS** *(n = 53 PwP, n = 50 SO, subsample self-reporting FM)*	**SUMMARY OF MIXED METHOD INFERENCES**
THEME Distancing and Disconnection **SUBTHEME & CODE (S)** Barrier to emotional engagement & connectedness	**a) FMQ-PwP Item-Level Responses**  **b) FMQ-SO Item-Level Responses**  	There was divergence of findings that FM alienates people with Parkinson's and their partners and family members. Items relevant to emotional distance on the FMQ were not as commonly or highly endorsed as highly as expected, given the interview findings. For SO, findings that FM may cause a sense of emotional distance converged.

Findings that SO perceived a decrease in emotional reciprocity partly converged (see figure 10, pg. 189). This consisted of two related findings, the loss of the ability to share in the moment, and a reduction in perceived emotional supportiveness of PwP. Interview findings suggested SO may perceive the inability of PwP to visibly display social and emotional understanding as signalling increasing emotional distance the within the relationship. Overall, FMQ item response distributions were consistent with these findings. Around half of SO endorsed item relating to experiences of feeling unsupported and reduced enjoyment or engagement in the relationship. Interestingly, SO's general perceptions of relationship quality and affection seemed to be relatively unaffected, with most SO respondents reporting they frequently experienced their relationship to be close and warm.

Qualitative findings revealed FM could result in a loss of perceived emotional reciprocity and was reported to cause distress and sadness for SO. Overall, the quantitative results converged with this finding. A substantial proportion of SO endorsed items referring to difficulty and alienation resulting from FM, which occurred at low to moderate frequency. Additionally, in Study 3, the greater degree of relationship difficulty related to FM (based on the whole group), the less marital satisfaction they reported and lower perceived availability of affectionate support (see Chapter 6). For all SO, an increase in FM-related difficulty also had significant links to decreased marital satisfaction and a lower perceived availability of affection support (see Chapter 6).

Figure 10

Joint display integrating the Qualitative and Quantitative findings on the reduction in emotional reciprocity within partnerships

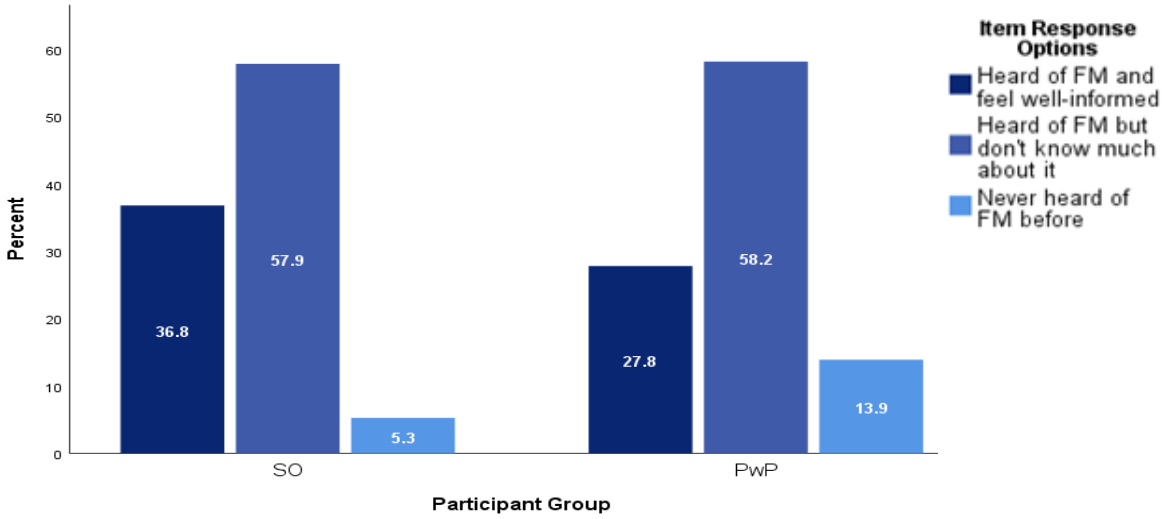
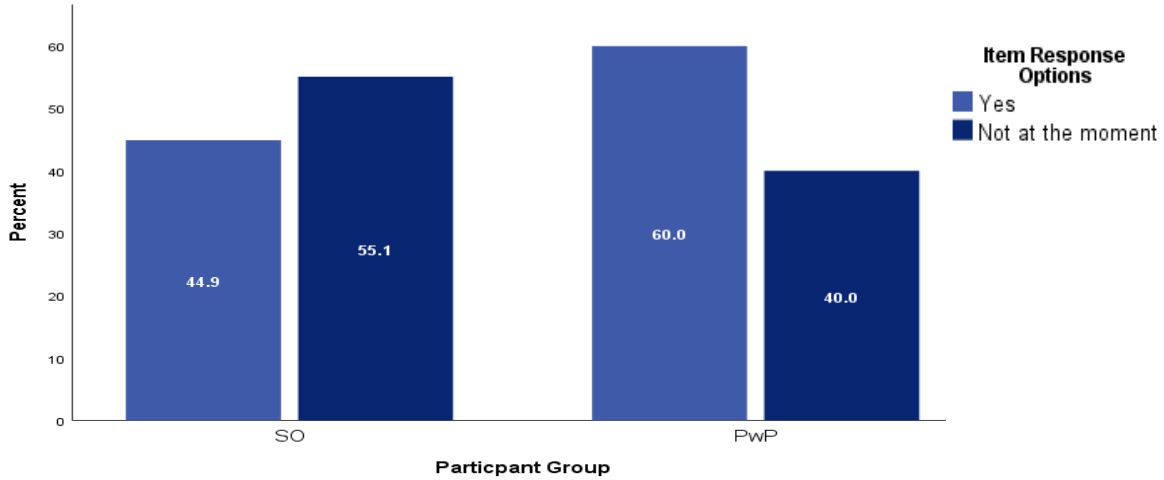
QUALITATIVE FINDINGS *(n = 9 SO)*	QUANTITATIVE FINDINGS *(n = 50 SO, subsample self-reporting FM)*	SUMMARY OF MIXED METHOD INFERENCES
THEME Decreased Emotional Reciprocity **SUBTHEME & CODE (S)** Loss of shared enjoyment & engagement Perceived supportiveness decreased Sadness at loss of (perceived) companionship		Findings that SOs perceived a decrease in emotional reciprocity partly converged. The finding that SOs experience distress or sadness at the lack of facial emotional responsiveness was confirmed. Over 70% of SOs reporting they struggled with the lack of response at least sometimes (see Figure 7b).

Findings that FM was a difficult symptom to recognise, and FM health education/ support were unmet needs converged (see figure 11, pg. 191). In the interviews, both PwP and SO reported FM was less conspicuous than some other aspects of PD, which made it difficult to recognise as a symptom of the condition. Interviewees also noted a lack of FM knowledge meant they less readily ascribed FM-related consequences in their relationships to PD. The finding that SO and PwP reported a generally low level of FM knowledge was confirmed, with a substantial majority of respondents endorsing that they knew either nothing or little about FM on the FMQ item pertaining to this topic.

Interview findings also indicated New Zealand PwP and SO desired greater access to FM-related information, with some individuals also wishing for greater dialogue with their healthcare providers about this topic. Responses on the FMQ item related to FM education/support needs were generally consistent with these findings. Most PwP respondents indicated a desire for more information about this topic on FM education items of the FMQ. A sizeable minority of SO also indicated they would like more FM health information or support on the FMQ.

Figure 11

Joint display integrating the Qualitative and Quantitative findings of FM health knowledge and education preference of the study

QUALITATIVE FINDINGS *(n = 9 PwP, n = 9 SO)*	QUANTITATIVE FINDINGS *(n = 53 PwP, n = 50 SO, subsample self-reporting FM)*	SUMMARY OF MIXED METHOD INFERENCES
THEMES An inconspicuous symptom Unmet health information needs **SUBTHEMES & CODES** Wish for greater access to FM education and/or support Importance of contextualising FM	**a) FMQ Item-Level Responses: Knowledge of FM at testing, by participant group**  **b) FMQ Item-Level Responses: Preference for further FM Education, by participant group**  	Findings that there was poor recognition of FM in PD, and that FM education was an unmet need converged. Findings that PwP and SO were found to desired greater access to FM-related information partly converged.

8. General Discussion

This thesis set out to study FM in the everyday lives of people with PD and their SO. The overarching aims of the research was to build a deeper understanding of FM from the viewpoint of those people who live with this symptom, and to investigate the potential association of FM with psychosocial wellbeing in PD. Thus, the research had two central aims:

1. To explore the lived experiences of people who have FM and their partners.

2. To assess the link between FM and psychosocial outcomes in PwP and SO.

The first aim was important to respond to gaps in existing literature by learning more about individual's and partner experiences of FM and their needs and preferences, so that FM could be better understood from the perspective of living with FM. Exploring personal experiences of FM also allowed for the identification of relevant concepts that could be developed into a clinical tool to support the management of the FM in PD. The second aim was important to build upon knowledge of the psychosocial consequences of FM as a general contribution to improving knowledge of PD.

The present section includes a general discussion of the main research insights and their application to clinical practice. This is followed by a broad statement on the strengths and limitations of the current programme of research and suggestions for future research directions. Finally, a general conclusion is presented.

What are the lived experiences of people who have FM and their partners?

There was great individual variation in the experience of FM for people who have PD and their family members. Common experiences that were described included the following: a lessened ability to read the person with PD; perceptions that the person with PD is in a persistent bad mood; jumping to conclusions about the lack of expression displayed; feelings of hurt or rejection; a loss of confidence in or concerns about the relationship; a sense of increasing disconnection or withdrawal within the relationship; distress at being unable to express/see the person inside; fears about future loss of facial expressivity; self-consciousness that FM will be negatively evaluated by others; and unkind reactions from others in casual interactions. Overall, the findings from the present programme of research and from existing studies (Gunnery et al., 2016; Ma et al., 2019) offer evidence that FM can have a detrimental impact on psychosocial wellbeing in PD. As scientific literature has scarcely begun to explore the psychosocial problems associated with FM or examined interventions to lessen their impact on quality of life, these findings speak to the need for greater research.

Psychosocial problems associated with FM also warrant greater clinical attention. FM is an aspect of PD that clinicians, their patients, and their families encounter nearly as regularly as speech deficits or tremor. Yet, most participants who self-reported FM in the present research described a difficulty identifying it, and poor knowledge of this symptom or how to cope to with it. Other PD populations may exhibit different characteristics. American's who had PD, for example, were found to have awareness of the level of expressive deficit (Mikos et al., 2009) and global self-ratings of the level of expressive difficulty have been shown to be significantly correlated with clinical evaluation of FM severity using the MDS-UPDRS, also in America (Ma et al., 2019). Nonetheless, it seems one key clinical undertaking for NZ PD populations may be improving awareness of FM and providing greater education to individuals with PD and their families.

This programme of research revealed novel insights of the ways FM is perceived, interpreted, and responded to, within the context of primary social relationships. Results suggested the impression bias commonly seen with unfamiliar observers also occurs in the context of primary social relationships. In familiar perceivers, the content, automaticity, and persistence of thoughts relating to (mis)perceptions of FM were revealed. FM was commonly perceived as a persistent bad mood or a lack of interest. SO frequently assumed they must be the cause of the perceived bad mood. A lack of perceived expressive response was also interpreted to signal discord in the relationship. Much like the evidence from healthcare professionals, some SO had a hard time overriding the negative impression bias FM creates, despite knowing FM was a symptom of PD.

One explanation for this negative impression bias is that the initial processing of emotion can occur automatically, often without intention or awareness of having done so (Lakin, 2006). In perceivers observing someone with PD, this could mean the lack of expression is automatically read as negative affect, even when perceivers know otherwise. In a second scenario, SO described a state of uncertainty because they were continually scrutinising the internal state of person with FM. This may be due to reduced, unclear, or conflicting emotional cues. Further description and investigation of these two scenarios is needed to better understand the ways FM is perceived in close social relationships and the various mechanisms which may underlie its misinterpretation. It may be that individuals who have greater FM, and therefore an absence of most or all expressive cues, are swiftly and globally judged by perceivers with a negative bias. It is possible when some degree of expression is retained an attenuation effect occurs, resulting in confusion for perceivers who attempt to make sense of weak, distorted, or conflicting expressive cues.

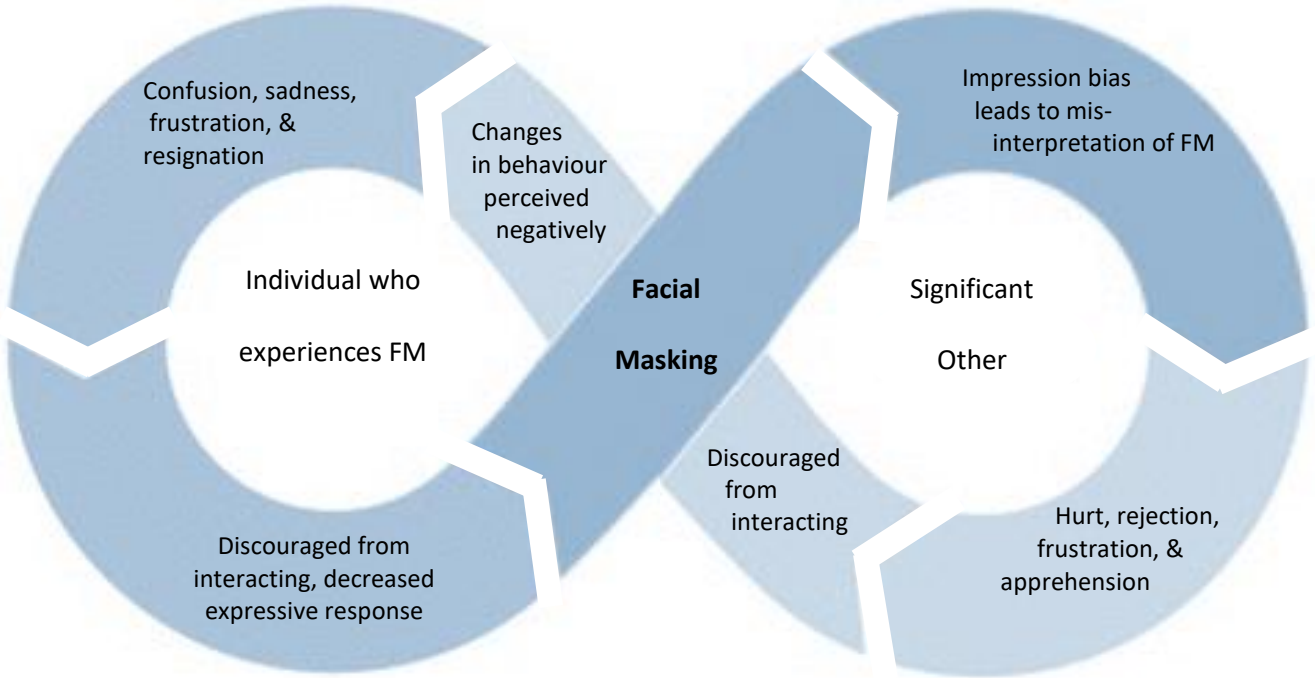
There is scare evidence of the way perceivers form impressions of individuals in other clinical conditions where facial movement and expression are impaired. It is also difficult to compare FM in PD to other clinical populations because the nature of expressive loss is different in each condition. In congenital facial paralysis (FP), individuals have greater opportunity to learn to adapt to living without facial expression and can take advantage of

others channels of communication (Bogart et al., 2014; Bogart et al., 2012), which often become impaired in PD, such as vocal and bodily expression. Individuals who acquire FP later in life are found to use fewer compensatory strategies than those with congenital FP (Bogart et al., 2014), but still have more options for adapting the communication of their thoughts and feelings than people with PD often do. Individuals with congenital FP are reported to describe experiences of social stigma and of being misunderstood by others, although the latter appear to be mainly associated with verbal communication being mistaken by others. (Bogart et al., 2012). Unlike in facial disfigurement or FP where irregularities in facial appearance are visible, the physical features of the face in FM appear largely unaltered, albeit abnormally still. The relative insidiousness of FM may be one explanation for the pervasiveness of the impression bias in FM, even in those perceivers who know the facial behaviours of an individual with FM are not valid cues.

Descriptions of how FM is misinterpreted are important because they can help us to better understand the effects of FM reported to occur within primary social relationships. As identified in this research and other qualitative studies (Chiong-Rivero et al., 2011; Schwartz et al., 2018), one such consequence was a growing emotional and interpersonal “gulf” within the relationships of people who have FM and their family members. Figure 12 depicts a proposed model for the dyadic processes that may be at work in FM, resulting in a pattern of increasing emotional alienation within close relationships. An adapted cognitive behavioural (CBT) framework can be used to understand this process (Beck, 1979; Padesky & Mooney, 1990; Teichman & Teichman, 1990; Teichman, 1992; Greenberger & Padesky, 1995; Grimmer, 2013). The process begins with the lack of expression being misinterpreted as a negative signal of internal state by sigficant others, such as emotional detachment or dissatisfaction. SO may also jump to inaccurate conclusions about the cause of what they perceive in the face of the individual with FM, often experiencing doubts about their relationship or blaming themselves for the perceived downturn in PwP mood or behaviour.

Figure 12

Proposed dyadic CBT process model depicting the pattern of misinterpretation and alienation that can occur due to FM in PD.



Note. Figure adapted from Psychology Tools (2020), *Reciprocal CBT Formulation*. Retrieved 31.08.2020 from https://www.psychologytools.com/articles/how-to-use-your-cbt-skills-to-conceptualize-relationship-and-interpersonal-problems-two-new-formulations-to-integrate-into-your-practice/

Naturally, such interpretations may cause emotional distress for SO, who commonly described feeling hurt, rejected, frustrated and apprehensive in response. SO could then become discouraged from interacting as frequently, warmly, or positively with the person who had FM.

At this point, the person with FM enters the cycle. The emotional and behavioural response of the SO is observed by the PwP, who also perceives these changes negatively. Feelings of sadness, confusion, resignation, and frustration were common in response. This could lead to increased social isolation and other changes in mood or behaviour for the PwP, which in turn, lessened opportunities for the social facilitation of expressive displays of emotion. The reciprocal misinterpretation and pattern of alienation results in fewer instances

of positive exchanges between the PwP and their SO, which would usually contribute to maintaining the emotional bond between them. In this way, both the PwP and SO become stuck in a cycle where they are both receiving inaccurate signals. These signals increasingly undermine the state of companionship, promoting feelings of doubt or self-blame, and increasing the sense of distance within the relationship. Initially this process may be triggered because sigficant others and PwP do not realise FM is a symptom of PD. While some participants from the qualitative investigation felt that gaining an understanding of FM broke this cycle, others had ongoing difficulty overriding misinterpretations of FM meaning the experience of a growing divide persisted. Further investigation and elaboration of this cycle is important because it could lead to the identification of areas for intervention (a topic further discussed below).

Similar models are proposed in facial difference/disfigurement literature. These theories incorporate the responses of other people to facial difference as well as the individual's own reactions, to explain psychosocial problems. In one model a negative feedback loop is described, where the unfavourable responses of others to the individual with facial disfigurement negatively influences the emotions and behaviour of that individual, in turn influencing or reinforcing the perceptions of others (Partridge, 1998; Clarke, 1999).The emergence of similar models that explain the consequence of facial expressive loss as an interaction between the individual and observers suggest it may be clinically important to incorporate partner or family members, to comprehensively and effectively address FM's psychosocial problems.

Evidence indicates healthcare practitioners understand some of the consequences of FM and recognise the importance of greater clinical communication about the psychosocial problems it can create (Schwartz et al., 2018). Taken together with findings from the present research, this suggests more could be done clinically about FM. While FM cannot currently be cured, there are potentially modifiable factors that could be targeted to lessen its impact, including greater awareness, early detection, and proactive health education. A key desire of participants who experienced FM in Study 1 was for greater education and dialogue with

their healthcare providers about his symptom. Unfortunately, some participants described a delay in recognising FM, which they felt detrimental to their wellbeing because the psychosocial consequences went unchecked. During recruitment for Studies 2 and 3, a sizeable proportion of PwP participants reported they suspected they had FM but were not sure, often stating this had not been addressed by their healthcare providers or discussed within their family. It is therefore advisable that healthcare practitioners routinely enquire about this symptom and offer education, regardless of the apparent clinical staging of FM seen on exam. Dialogues which assist individuals and their families to recognise the issues potentially explained by FM and identify possible coping strategies could help people mitigate the cycle of increasing interpersonal disconnection that can occur (see Figure 13). The FMQ (developed in Study 2) is one clinical tool that could increase awareness in all parties in ways they may not be thinking of, and form part of the patient education process in understanding FM.

In summary, the negativity bias associated with stranger and healthcare professional perceptions also occurs in the context of relationships partners or close family members in PD. The influence of FM in close relationships extends beyond the inaccurate social judgements seen in less familiar receivers. In close relationships, FM leads to false perceptions that may promote emotional detachment and a loss of confidence or security within the relationship. The reciprocal cycle of misinterpretations and alienation that may occur within close relationships in FM, could be particularly damaging when FM remains unattributed to disease-related changes. Increasing awareness, education, and early detection are clinically important in managing FM. Greater clinical attention and proactive healthcare communication about FM could help mitigate the emotional and interpersonal gulf that can occur in the relationships of people who have PD.

What is the link between FM and psychosocial outcomes in PD?

Overall, the more negative consequences of FM that SO reported, the lower the level of marital satisfaction, more caregiver strain, the less available they perceived affectionate support or positive social interaction, and the worse psychological health they experienced. In PwP, the more negative consequences of FM that participants reported, the more the anxiety and depression symptoms that they experienced. They also experienced lower levels of marital satisfaction, perceived a lower availability of affectionate support and a lower availability of positive social interactions. The link between the latter social relationship variables with FM in PwP, however, remains unclear. Not all relationship difficulty items on the FMQ explicitly attributed problems to FM, so may be measuring aspects of general relationship functioning. Answers to those FMQ items with ambiguous wording might reflect general relationship impressions predating FM, rather than the impact of facial expressive loss.

Also, PwP self-report of FM severity was not found to be linked PwP perceptions of FM impact or other social relationship outcomes measured. A possible explanation for this is a differential impact of FM for PwP and SO. Part of this explanation may be that PwP do not recognise the impact of FM in their relationships, even if FM is having an effect on their partner and (potentially) ultimately on PwP marital satisfaction or relationship functioning. This is consistent with the idea proposed by Gunnery and colleagues (2016), that the relationship perceptions of people who have PD may be sheltered from the influence of FM. The clinical implications of this are that partners' experiences of FM should be included where possible, to help enhance understanding of FM in both individuals and their clinicians. Evidence evaluating successful outcomes in psychological interventions for people with PD is also supportive of the involvement of SO. Caregiver participation in CBT for PwP depression has been found to be a significant predictor of a positive treatment response (Dobkin et al., 2012).

There were significant associations between participants' perceptions of FM severity

and the amount of FM-related difficulty they experienced. Interestingly, no significant correlations were found between healthcare professionals' clinical ratings of FM and participants ratings of FM severity. Moreover, there was no correlation between healthcare professional ratings of FM and nearly all health and wellbeing outcomes measured (except for symptoms of anxiety, which had a small negative correlation with healthcare professional ratings). This is in line with previous studies that found the expressivity of people with PD varied when in recorded in conversation with their spouse, compared to conversations with a researcher (Simons et al., 2004). It is also possible this could also be due to methodological reasons, such as the inclusion of only two healthcare professionals who were unfamiliar with most of the PwP in the present research. The usual/treating healthcare practitioner of participants with PD might provide different ratings of FM severity. It also possible that the level of FM is perceived differently by healthcare professionals, who assess for specific clinical features of FM. The implication of this finding for practice is that relying solely on clinical evaluations of FM may not provide sufficient assessment of the possible effects of this symptom for patients and their families.

The present research also provided an initial description of the ways PwP and SO adjusted to FM. Compensatory behaviours and coping strategies described by participants could be categorised into two broad approaches: practical and psychological. Practical strategies included enhancing communication and rehearsing expressions to promote greater facial mobility. One notable finding was that posing expressions (making a socially appropriate smile) was regarded as too effortful and contrived for many of the participants with PD. Healthcare professionals should be aware of this preference when providing information or support to individuals with FM. While people with FM may have the ability to use this strategy, it might not a practical or realistic approach to managing FM for some individuals. Psychologically based approaches included challenging false interpretations of FM (for SO), having an accepting and positive mental outlook, and various relationship strengthening behaviours. Methods for SO to manage unhelpful misinterpretations of FM seem like an important area for future research, because they are a common occurrence

and seem to be difficult to override. There is some indication in the broader literature on facial expressive impairment that training programmes for the perceiver may reduce the impression bias. Training perceivers to focus on the voice and body of people with facial paralysis was found to improve bias in impressions of extraversion but did not improve the accuracy of perceivers' impressions (Bogart & Tickle-Degnen, 2015). Also, if models integrating the interpersonal and psychological in FM are accurate, then SO perceptions and experiences of FM play a significant role in the impact of this symptom for both persons in a relationship.

Although the link between FM and psychosocial wellbeing in PD seems to have received greater attention recently, current literature is characterised by a lack publications on psychosocial intervention and clinical management for FM in PD. FM interventions in published literature are impairment-oriented and focused on retaining or improving physical function of facial musculature (Katsikitis & Pilowsky, 1991; Dumer et al., 2014; Ricciardi et al., 2016). While physical mobility clearly plays an important role in facial functions, this neglects other issues and concerns encountered by individuals who have FM, including the emotional and interpersonal. Much of published research on psychological therapies in PD has focused on CBT for the treatment of depression (Spencer & Haub, 2018; Zarotti et al., 2020). No published studies could be located that take a psychosocial approach to treating FM in PD. One UK-based community organisation, Changing Faces, uses a CBT- based self-model to help people cope with facial disfigurement (FD), with an overall aim of improving self-esteem and body-image (Partridge, 1998; Clarke, 1999). In Clarke (1999), the key components are reported as exploring negative feelings about losing looks, challenging unrealistic beliefs about possible treatment or unhelpful beliefs about the future with FD, and learning problem-focused coping skills. A different CBT outpatient programme for facial and bodily disfigurement evaluated post treatment outcomes, finding social anxiety, depression, and distress related to appearance were significantly improved (Kleve et al., 2002). CBT could be adapted to focus on key problems of FM and tested in PD. Future research could also explore the potential benefits of adapting other existing therapies that

treat psychological and social issues in people with PD, such as family systems therapy (Spencer & Haub, 2018). Topics which could be usefully explored in psychological interventions for FM could include: intense anxiety about the future personal consequences of developing FM; recognising and challenging negative cognitions that stem from the misinterpretation of FM (the impression bias); problem-solving for FM-related difficulties that cause relationship discord.

In summary, FM does have a detrimental effect on social and psychological wellbeing in PD but may have a differential impact for PwP and their SO. It was participants perceptions of the impact of FM-related difficulties, rather than symptom severity per se, which was most strongly linked to poorer psychosocial outcomes. People who experienced FM used a range of strategies to help them cope but still desired greater education and support around this symptom. FM targeted psychosocial interventions are yet to be published in the literature, however existing psychological therapies in PD could be adapted to focus on FM-related difficulties.

Strengths, Limitations & Directions for Future Research

There are limitations with respect to the participant cohorts of this research. One limitation was the sample size, the level of FM severity, and range of PD stages represented. While a handful of people with a higher degree of FM took part, future research would benefit from including a greater proportion of people with severe FM. Another limitation was the recruitment of research participants who mostly came from Parkinson's support group organisations, due to the lack of a central PD register within the New Zealand public health system. There may be differences in the characteristics of people who choose to engage in support services compared to those who do not. Self-selection of individuals who are relatively free of psychosocial stressors and/or at an earlier stage of PD might explain the generally positive findings on mental health and wellbeing measure for PwP in the present research. These limitations are in part a reflection of the relatively small NZ population and therefore small number of people PD (and FM) in NZ. They also reflect the lack of regular PD clinic times within the NZ public system. Replication of this research with a large multi-site or international recruitment strategy would help to clarify the findings, in particular further validation of the FMQ (Study 2) is recommended.

From a methodological perspective, the interpretations of the findings of Study 3 were limited because of the use of different methods of assessment and measurement instruments. Assessment of self-reported FM relied on a new questionnaire (the FMQ) in PwP and their SO, as no other patient or partner report measures could be located. The FMQ has good reliability and initial evidence of validity but may not accurately capture FM and its impact. Other FM studies have used a global rating item asking participants to rate the amount of expressivity difficulty they perceive on a 5-point scale, where 1 = no difficulty and 5 = severe difficulty (Gunnery et al., 2016; Ma et al., 2019). There may be differences in the characteristics of FM captured or emphasised by various methods of FM measurement. Due to practical restrictions, healthcare professionals used established clinical staging tools while viewing video recordings of participants, rather than in vivo observation with patient's

usual clinicians, which may have provided different ratings of FM severity. Further to this point, only two healthcare professionals provided assessment of FM severity, and those who took part were familiar with but not greatly experienced in using MDS-UPDRS rating scale.

Findings from the present programme of research suggest a number of additional directions for future investigations (other than those outlined above). Besides the main issue that outcomes of FM for patients and their families have been generally neglected in PD literature, there is a need for more comprehensive investigation of the differential impact of FM for PwP compared to SO. It could also be beneficial to examine the factors that could belie good adjustment to FM, such as psychological flexibility or the possible interaction between PD onset type and/or FM severity with the ability to meet social role demands across different life stages. This research began to describe the compensatory methods used by people with FM and their partners but did not investigate what strategies work best or for whom, which would be an interesting topic for future research. The focus of the present project and existing FM research has been the consequences for primary social relationships, the influence on interactions with healthcare professionals, and the influence on interactions with strangers. Future research could describe the influence of FM in relationships with other key members of support networks for people with PD, including support/rehabilitation programme associates, cultural or community groups, work colleagues, and wider family or friends. Different factors may influence psychosocial wellbeing across various types of social relationships, and this could be further investigated. Lastly, it was beyond the scope of this thesis to investigate the link between FM and changes to speech, and with other types of affective dysregulation that co-occur in PD. Current research has generally focused on individual variables such as FM, emotion recognition, or speech difficulties. It might be beneficial to take a holistic approach to understanding how these deficits affect socioemotional functioning in PD.

Conclusion

This thesis set out to understand the lived experiences and psychosocial consequences of FM for PwP and their SO. Findings from the initial qualitative investigation concluded FM does interfere with the ability of SO to decode the internal state of the person who has PD and can have a negative effect on subjective wellbeing for individuals and couples. Study 2 presented the development and validation of two targeted questionnaires which could help to improve clinical assessment of FM impact. The results of study 3 adds to what is known about the implications of FM, indicating that greater perceived FM severity and/or higher levels of difficulty related to FM are associated with poorer outcomes in some aspects of social and psychological wellbeing.

Overall, the findings from the present programme of research and existing evidence suggest FM can have a detrimental impact on psychosocial wellbeing in PD. However, FM may influence people who have PD and their partners/family members differently. More comprehensive exploration is required to better understand the psychosocial outcomes of FM in primary social relationships with close friends and family. As FM does have a negative impact for some people with PD and their family members, it warrants greater clinical attention. Therefore, two key directions for future research may include the adaptation and evaluation of psychological therapies for FM-related difficulty and investigation of the factors associated with good adjustment to this unique aspect of PD.

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Appendices

Co-authorship forms

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Co-Authorship Form

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Please indicate the chapter/section/pages of this thesis that are extracted from a co-authored work and give the title and publication details or details of submission of the co-authored work.

Thesis Chapter 4, Study 1, pp. 39-52
"Unmoving and Unmoved, Experiences and Consequences of impaired non-verbal expressivity in Parkinson's patients and their spouses"
Published in Disability and Rehabilitation, 41:21, pp. 2516-2527. DOI: 10.1080/09638288.2018.1471166

Nature of contribution by PhD candidate	Adrienne Wootton
Extent of contribution by PhD candidate (%)	80

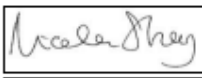
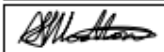
CO-AUTHORS

Name	Nature of Contribution
Professor Nicola Starkey	Analysis, feedback on drafts of paper and responses to reviewers comments
Dr Carrie Barber	Consultation on design and analysis, and feedback on drafts
Adrienne Wootton	Conceptualisation of the study, ethics approval, data collection and analysis, main author of paper.

Certification by Co-Authors

The undersigned hereby certify that:

- the above statement correctly reflects the nature and extent of the PhD candidate's contribution to this work, and the nature of the contribution of each of the co-authors; and

Name	Signature	Date
Professor Nicola Starkey		12.01.21
Dr Carrie Barber		13.1.21
Adrienne Wootton		12.1.21

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Nature of contribution by PhD candidate	Adrienne Wootton
Extent of contribution by PhD candidate (%)	70

CO-AUTHORS

Name	Nature of Contribution
Professor Nicola Starkey	Advice on analysis, input and feedback on drafts of the manuscript
Dr Carrie Barber	Consultation on design and analysis, and feedback on drafts
Adrienne Wootton	Conceptualisation of the study, ethics approval, data collection and analysis, main author of paper.

Certification by Co-Authors

The undersigned hereby certify that:

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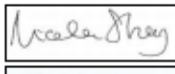
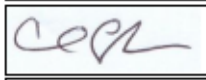
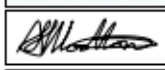
Professor Nicola Starkey		12.01.21
Dr Carrie Barber		13/1/21
Adrienne Wootton		12.1.21

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Please indicate the chapter/section/pages of this thesis that are extracted from a co-authored work and give the title and publication details or details of submission of the co-authored work. Thesis Chapter 6, Study 3, pp. 132-172. "Face Value: Perception of Facial Masking and the link to Psychosocial Outcomes in Parkinson's Disease" Prepared for submission	
Nature of contribution by PhD candidate	Adrienne Wootton
Extent of contribution by PhD candidate (%)	70

CO-AUTHORS

Name	Nature of Contribution
Professor Nicola Starkey	Study design, analysis, providing feedback on drafts of paper
Dr Carrie Barber	Consultation on design and analysis, and feedback on drafts
Adrienne Wootton	Conceptualisation of the study, ethics, data collection and analysis, main author of paper.

Certification by Co-Authors

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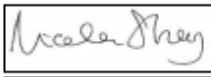
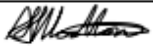
Professor Nicola Starkey		12.01.21
Dr Carol C. Barber		13/1/21
Adrienne Wootton		12.1.21

Copy of Parkinson's New Zealand article and revised fact sheet

THE IMPACT OF FACIAL MASKING

With Adrienne Wootton

The Parkinsonian was pleased to spend time speaking with Adrienne Wootton recently about her work.

WAIKATO UNIVERSITY CLINICAL PSYCHOLOGY PHD CANDIDATE, ADRIENNE WOOTTON.

It was an acquaintance with Parkinson's losing her ability to smile that led Waikato University PhD student Adrienne Wootton to study facial masking in Parkinson's.

Adrienne, 28, was already studying clinical psychology. In her words, she had "always been fascinated by mind, brain and behaviour relationships."

"Over the course of my studies in psychology and training in clinical psychology, I developed a particular interest in neuropsychology and conditions such as dementia and Parkinson's.

"[When the woman she knew lost her ability to smile] I looked into the scientific literature and found very few studies had investigated the impact or management of facial masking. It struck me that maybe this symptom could interfere with patients and their family's mental wellbeing and quality of life. This idea became the basis for my present research."

Now Adrienne, a Hamiltonian who completed her undergraduate studies at Auckland University, is back at Waikato, halfway through a thesis on the psycho-social aspect of facial masking.

She recently completed a qualitative study on people with Parkinson's who have facial masking and how masking affects their relationships with their life partners.

Adrienne says facial masking affects some people with Parkinson's, who may have a reduced capacity to move their facial muscles and display expressions. This is caused by lower than normal levels of the neurotransmitter, dopamine, in their brains that also impacts on a person's ability to walk, run, move their hands and carry out other co-ordinated movements.

Adrienne's study found that many people with facial masking did not initially realise they had it.

Interestingly, it also suggested that partners of people who have Parkinson's and facial masking were more directly affected by it than the people themselves.

Adrienne interviewed nine people with Parkinson's who had facial masking and their partners (18 people in total).

A third of participants reported substantial negative effects on their own well-being and/or their relationship with their partner because of facial masking, another third reported some effects and a third segment reported minimal or no effect.

Despite the significance of facial masking, many people with it only found out through being asked to smile for a photograph, for example, when they thought they were already smiling. Or when other people commented that they never smiled or seemed grumpy all the time.

Another key finding was that partners commonly misidentified facial masking as negative emotions. Addressing the issue of "blank" expressions, one participant commented if you didn't know how a person was feeling, you tended to fill in the gaps with negatives such as anger, sadness and similar. Partners worried about why the person with masking might look this way, for example, thinking they must be unhappy with the relationship. These (often incorrect) assumptions could cause partners to feel anxiety or distress. The study also emphasised such looks were not just "neutral" but could involve the mouth of the patient being open, their jaw dropped and less blinking than normal.

It was essential facial masking was identified and considered for what it was. On a more positive note, the study found that once people with masking and their partners became aware of how masking might make a false impression; this could make a significant difference in helping them deal with the effects.

Adrienne said the present research suggests negative consequences of masking, such as relationship strain and feelings of isolation or distress, may be unnecessarily prolonged, if the patient and their family remained unaware of this symptom.

Masking could become a barrier to physical and emotional intimacy. Around one half of the participants reported feelings of distance and disconnection in their relationships with most attributing it to facial masking all or in part. There was sometimes a self-perpetuating aspect. The partner would perhaps feel hurt or resentful about the person with facial masking's apparent coolness, which would make the person feel more isolated, and so on.

"One person with facial masking likened it to being in a dark room with their partner not seeing them but only hearing their voice."

Speaking on the significance of the study, Adrienne told the *Parkinsonian*: "Emotional and social consequences are not always included in the clinical assessment and management of Parkinson's, but are often important to patients and their families. Psychological and social wellbeing are especially important because of the long term nature of the condition and long duration of care usually provided by family members."

Adrienne has benefitted from support from and through the Bryant Trust Postgraduate Research Scholarship and the University of Waikato Doctoral Scholarship.

FACT SHEET

What is facial masking?

Facial masking is a problem for some people with Parkinson's

WHAT IS FACIAL MASKING?

There are 43 muscles in the human face that enable us to smile, frown and express subtle feelings. It is the lack of the neurotransmitter, dopamine, in the Parkinson's brain that diminishes people's ability to walk, run, move their hands and carry out other movements in a co-ordinated way. It also sometimes also reduces the ability to control facial movements and expressions. This is called facial masking.

IS FACIAL MASKING SOMETHING YOU EXPERIENCE IF YOU HAVE PARKINSON'S?

No, like other Parkinson's symptoms, facial masking is a problem for some people and not others.

WHAT DOES FACIAL MASKING LOOK LIKE?

This very much depends on the person. Expressions like a smile may look "watered down" or your face may show little expression at all. Other key signs include a reduction in blinking which may give an appearance of staring, the jaw dropping open when your mouth is at rest, and an overall look of being stone-faced or flat.

WHAT CHALLENGES DOES FACIAL MASKING PRESENT?

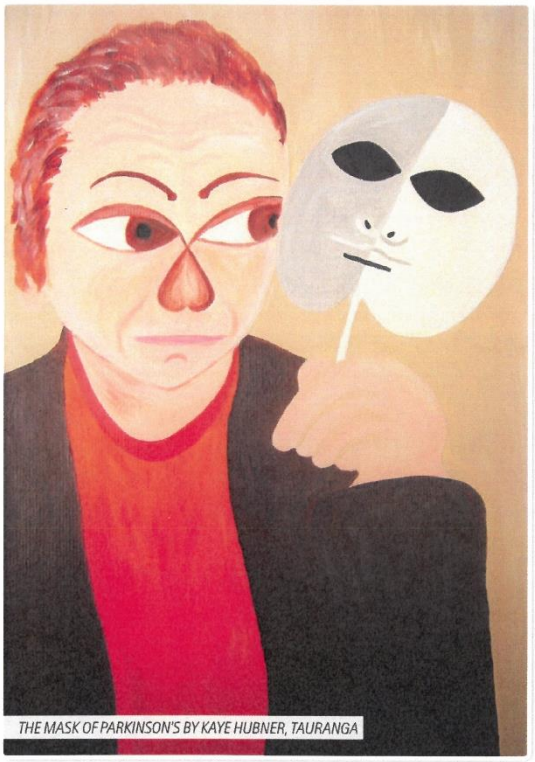
Maintaining satisfying relationships is an essential component of wellbeing, with our faces being a fundamental part of how we connect with other people. If you have problems with facial masking, communicating, socialising, and bonding with others can be more of a challenge.

Although facial masking is a common sign of Parkinson's, there is a lot of variation in how noticeable different people find this symptom and the impact it has for them. Some people find it negatively affects their relationships and sense of mental wellbeing. Others report masking does not trouble them. There is ongoing research into how we can better understand facial masking and ways to live well with it.

"When there's little expression to read, you fill in the gaps"

One of the main issues with masking is that when there is less expression to read, other people tend to fill in the gaps. Often, masking is misread as a bad mood. Sometimes, other people interpret masking as a lack of care or lack of interest. You may feel good and think you are smiling at someone, for example, but facial masking means others do not see it and assume something is wrong. People who know you well may react in unusual ways, such as asking you if are grumpy, apologising, or behaving in a more reserved way around you.

The way other people see masking can be difficult to override. If you are the partner of someone with masking, you might find yourself second guessing what your loved one is thinking, feeling, or saying. It is common to feel apprehensive or discouraged at the lack of expression. You might even feel like the look on the person with masking's face is your fault in some way.



THE MASK OF PARKINSON'S BY KAYE HUBNER, TAURANGA

The way masking is interpreted can be particularly problematic when people don't realise it is part of Parkinson's, and wrongly make assumptions based on how the face looks. Many partners find it is helpful to remind themselves what is seen on the outside doesn't match how the person feels on the inside (for more tips on managing masking, see below).

"It can put a couple apart"

Masking can make it harder to share in the moment with friends and family. This can contribute to feelings of being isolated from the people who are important to you, including your partner. Your loved ones might also feel as though you have grown distant or detached.

Sometimes both people might feel as though the other is pulling away, when this is not the intention of either person at all. This occurs because other people misinterpret masking, and you may misinterpret other people's reactions to your facial appearance (and so on). This cycle can be broken or minimised by coming to realise masking is a symptom of Parkinson's, rather than a reflection of feeling.

MANAGING THE CHALLENGES OF FACIAL MASKING

First of all, there are things people can do to help manage masking. Whether it is you, your partner, or your health professional, it's essential to recognise masking as a part of Parkinson's and come to terms with the fact you/your loved one may come across differently to how they think and feel inside. Many people find this helped to put things in perspective.

People with facial masking and their carers have reported several things are helpful in managing this symptom (see below). Just as every person who has Parkinson's is unique, so is every person who experiences facial masking. What works well for one person might not be suited to another. It's important to find what works best for you and your family.

MANAGING FACIAL MASKING

People with facial masking and their carers have reported a number of things are helpful in managing this symptom:

If you experience facial masking:

- **Say what you feel.** Speaking aloud thoughts or describing how you feel can help others see past masking.
- **Undertake physical exercise for the body and face**. There is some evidence it can help persevere the ability to express yourself. This can be an organised program or regular practice of moving your facial muscles at home.
- **Replace a smile with touch or speech**. Finding other ways to show you care helps maintain your connections with loved ones.
- **Make a conscious effort to smile.** Some people find this works well for them; others prefer not to do this.

If your loved one has facial masking:

- **Be mindful** the person may look to be in a bad mood, but likely doesn't feel that way inside.
- **Ask questions or agree to check in.** Having a conversation helps to prevent misunderstandings and worries. Agreeing to check in regularly can also be a good compromise.
- **Catch out your reactions to facial masking.** It is common to feel discouraged at the lack of expression. Recognise when this happens and rely on what the person says and does, rather than how their face looks.
- **Reconnect to the person inside**. Having a reminder of the smile and spirit inside can help. This could be looking at a cherished photograph or recalling a happy memory.

You and your partner can both:

- **Tell close friends and family** about facial masking. It can make it easier when everyone understands and realises what is happening.
- **Take time out together and be kind** to one another. This can be good for any partnership where there are stresses or strains.

Source: The University of Waikato Parkinson's Facial Masking Doctoral Research Project, Parkinson's UK